

# Evaluating the 'Parental Understanding of Neurodisability Questionnaire' as a measure of change

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## **ABSTRACT**

This thesis aimed to provide a second validation phase of a recently developed measure: the Parental Understanding of Neurodisability Questionnaire (PUN-Q) (Moran et al., submitted). The PUN-Q is a thirteen item self-report questionnaire measuring parents' understanding of their child's neurodisability. This thesis prospectively validated the PUN-Q over three time points, prior to and following a child's attendance at a Tier-Four paediatric diagnostic assessment, for queries regarding social communication. Four main aims were investigated: 1) to establish prospective Construct Validity by comparing the PUN-Q to two other parent-report measures (perceived self-efficacy and parenting stress); 2) to examine test-retest reliability of the PUN-Q by comparing two pre-assessment time points; 3) to examine whether the PUN-Q is sensitive measuring potential pre-and-post assessment changes to parental understanding; 4) to explore the relationship between the PUN-Q and child emotional, behavioural and social communication difficulties. These objectives were addressed using data collected from 37 parents, due to time constraints the study was underpowered at Times two and three (n=26, n=11, respectively); bootstrapping confidence intervals were therefore estimated for non-parametric data. Evidence was provided for construct validity at Time 1, but not at Time 3. Test-retest reliability was suggested for the PUN-Q between two non-intervention time points. Results suggested that the PUN-Q is responsive to changes over time, and that the clinic's diagnostic assessment is effective in enhancing parental understanding. The PUN-Q was not shown to be related to child-related outcomes. These preliminary results suggest that the PUN-Q is an important measure that can reliably and conveniently measure parental understanding of their child's neurodisability symptoms. This study suggests a role for parental understanding within a wider model of parenting stress and coping with disability. Further validation is needed to allow dissemination to the wider neurodisability service, and to less complex symptom presentations.

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## **CHAPTER 1: INTRODUCTION**

### **1.1 General Overview**

This thesis aimed to provide further validation for a newly developed measure: the ‘Parental Understanding of Neurodisability Questionnaire’ (PUN-Q). The PUN-Q is a parent self-report tool, which measures parents’ perceived understanding of their child’s neurodisability symptoms, the unique impact that these symptoms may have upon their child, and their understanding of their child’s developmental needs and management. The PUN-Q is the only identified instrument within the literature to systematically measure parental understanding within the context of neurodisability. This aspect is important to consider within clinical assessment settings, as parents are often required to deliver interventions and advocate for their child’s developmental needs to be met. Evaluating parental understanding may therefore be essential to help maximise the effectiveness of any subsequent interventions (e.g. Ho et al., 1994).

The PUN-Q was developed and initially validated using a sample of parents whose children had suspected Autism Spectrum Disorder (ASD). Parents were recruited within a Tier-Four paediatric neurodisability service in London (Moran, Pote, Topper, & Dale, submitted). The current study was set up as a second validation stage, with an independent sample of parents. In order to ensure continuity between the two studies, the current study also recruited parents whose children were referred to the clinic for queries regarding ASD. Novel to this study was its aim to assess the PUN-Q’s stability over time (i.e. test-retest reliability), in addition to its sensitivity to detect any changes to parental understanding, following the administration of a multi-disciplinary neurodisability assessment. Accordingly, this thesis aimed to provide a preliminary examination of whether a multi-disciplinary diagnostic assessment can help to improve parental understanding within this selected group.



This thesis also aimed to extend previous construct validation of the PUN-Q, by examining if it was concurrently related to previously identified parental covariates: parenting stress and perceived parental self-efficacy, before and after the neurodisability assessment. The final novel aim of this thesis was to learn more about the workings of the PUN-Q and parental understanding of neurodisability as a concept, by investigating how the PUN-Q related to two child measures for emotional, behavioural and social communication difficulties.

The following sections will review the literature, outlining the rationale for the PUN-Q measurement of parental understanding and its importance within the context of neurodisability. The review will focus specifically on ASD and the process of diagnosis. Previous empirical research will be included which has investigated parent related outcomes: parenting stress levels and self-efficacy beliefs, suggesting how these might relate to parental understanding. Finally, this chapter will outline the hypotheses and aims for the current study.

As aforementioned, the current study will focus only on the neurodevelopmental disorder of ASD. However, due to the paucity of published research focusing specifically on parental understanding of child neurodisability, where necessary and appropriate the following review will draw on relevant empirical findings from a wider range of neurodisabilities reported on within the literature.

## **1.2 Definition of Parental Understanding of their Child's Neurodisability**

For the purposes of this thesis, parental understanding is defined as the manner in which parents understand their child's suspected or diagnosed neurodisability symptoms, and the unique impact that these symptoms may have upon their child. No identified study has systematically measured parental understanding within the context of neurodisability and

through the use of a scientifically validated tool. Other authors have written about similar or related concepts such as: ‘Parental Awareness’ for typically developing children (e.g. Newberger, 1980), ‘parental cognitions’ regarding children’s disability (e.g. Hassall & Rose, 2005), or the level of parent-professional agreement (e.g. Geiger, 2002). These previous concepts are not thought to wholly encapsulate parental understanding as measured by the PUN-Q, which is specific to child neurodisability. They will be outlined within the following section to provide context for the development of the PUN-Q as a needed measure of parental understanding.

It is hypothesised here that parental understanding is multi-faceted, incorporating different cognitions that parents may have regarding their child, for example, appraisals, meanings attached to salient events or beliefs about themselves and their child (Hassall & Rose, 2005). Parental understanding also refers to parents beliefs regarding the impact that a potential or undiagnosed disability may have upon their child; i.e. the individual and idiosyncratic profile, prognosis and consequences of their child’s symptoms (Moran et al., submitted).

Historically, Newberger (1980) proposes parental understanding to be important in enhancing the development of healthy children. According to her postulations, ‘Parental Awareness’ should adapt according to a child’s developmental level. This adaptation enables parents to express appropriate empathy for their child’s needs and enhance parent-child interactions (Newberger, 1980). ‘Parental Awareness’ potentially overlaps with parental understanding of child neurodisability, however, this model is based on typical child development and may not therefore be fully relevant. Newberger’s (1980) model can provide context for parental understanding in neurodisability by demonstrating the difficulties that such parents may have in developing deeper understanding of their child.

‘Parental Awareness’ for typically developing children is proposed to consist of four hierarchical levels (Newberger, 1980): 1) ‘*Egoistic understanding*’ of the child based on a parents’ own needs; 2) ‘*Conventional understanding*’ of the child, with the parental role based on environmental factors (e.g. culture, tradition, or knowledge of child development); 3) ‘*Subjective Individualistic Understanding*’ of the child as a separate person. Within this level, the parental role is specific to the child’s unique needs, and not based on societal norms or stipulations; 4) ‘*Interactional Understanding*’ of the complex and changing nature of the child and their needs based on an expected developmental trajectory. At this highest level of awareness parents are able to understand and balance a child’s needs with their own, in order to form a healthy interactional relationship. This final stage of awareness is perhaps closest to the level of parental understanding that may be needed for parents in caring for their child within the context of neurodisability.

At each of these four levels, parents are increasingly able to understand the impact of the environment and of their own parenting strategies upon their child. This changing and growing awareness allows parents to formulate parenting strategies appropriate to their child’s developmental abilities (Newberger, 1980). This model is however based on the premise that parents’ cognitions regarding their child will be stable across different aspects of parenting. This may not be the case for ASD, which is a highly heterogeneous disorder: symptoms can vary in severity between children, at different stages of development and across life-skills (Baird et al., 2008). Newberger’s (1980) model does however provide a possible developmental framework to anticipate the levels of parental understanding that may be needed in order for parents to feel able to meet their child’s needs, within the context of neurodisability.

Parents of children with neurodisability symptoms may need to develop all four levels of ‘Parental Awareness’ in order to gain an adequate level of understanding. For example,

*'conventional understanding'* may reflect disorder specific knowledge, which can be gained through liaison with health-care professionals or independent research. Parental awareness of neurodisability, which comprises disorder specific knowledge, reflects understanding that is based on information about the overall population with that specific disorder/ disability. This form of understanding is not therefore individualised to the specific child.

Parental understanding within the context of neurodisability should be applicable to a specific child, incorporating parents' perceptions of the *impact* that a disorder or set of symptoms may have upon *their* child and the process by which they can shape expectations for the future (e.g. prognoses) (see Dale, 1996). A higher level of understanding is therefore gained through subjective or *'individualistic understanding'*, which enables parents to appreciate how neurodevelopmental disorders and symptoms may uniquely affect their child. This allows the child to be understood both within the context of disorder specific knowledge, in addition to the *individualistic impact* of the disorder.

The developmental trajectory for children with complex neurodevelopmental needs can change unpredictably (Hewitt-Taylor, 2005), which may make it difficult for parents to attain the highest levels of awareness. Difficulties developing adequate awareness of their child can cause parents to feel 'powerless' and unable to fully participate in their child's treatment (Dale, 1996). Specialist clinical assessments aim firstly to clarify the diagnosis and secondly to help parents understand how a diagnosis fits their child. This process can potentially help to enhance parents' overall understanding of their child (Mittal, Sciberras, Sewell, and Efron, 2014). In order to evaluate the effectiveness of such input, the use of an outcome or screening measure which is sensitive and specific to changing levels of parental understanding is therefore desirable; the PUN-Q was developed for this purpose and has so far been validated retrospectively on parents of children with complex ASD symptoms (Moran et al., submitted). No other measure of this kind has been identified within the literature.

As aforementioned, parental understanding of a child's neurodisability can also be considered through examining parental cognitions including health-related concepts such as 'Health Literacy'; as defined by the World Health Organisation (Nutbeam, 1998). Levels of 'Health Literacy' determine an individual's cognitive and social skills, which enable them to access, understand and utilise information, in order to '*promote and maintain good health*' (Nutbeam, 1998). Within the context of paediatric services, this will reflect the extent to which parents are able to comprehend and utilise information provided by health-care professionals. Difficulties gaining parental understanding have been shown with regards to paediatric congenital heart disease. Approximately half of the 156 parents surveyed were unaware of the possible aetiologies and symptoms associated with their child's disorder (Cheuk, Wong, Choi, Chau, and Cheung, 2004).

It is important to identify parents' understanding of healthcare input, as parents' health-related cognitions (i.e. beliefs and attitudes about their child's health or about themselves) can influence the development of specific parenting strategies and thereby impact on children's health outcomes (see review by Bugental and Johnston, 2000). A survey conducted with 77 parents of children with mild Learning Disabilities showed that parental understanding and acceptance of their child's special health-care needs was associated with parents' adherence to treatment recommendations, following a psycho-education based assessment (Human & Teglas, 1993). However, within this study, maternal IQ was positively associated with participant attrition, suggesting a possible bias within the final sample. It is also unclear whether these results would generalise to children with more complex neurodisability symptoms. Taking these limitations into account, these results suggest that parental cognitions are important in helping parents to develop positive and appropriate coping strategies (e.g. planning or problem solving), which can affect parents' abilities to develop appropriate parenting strategies (see Cunningham & Davis, 1985; Dale, 1996).

Within their review of studies investigating parental cognition and children's intellectual disability, Hassall and Rose (2005) suggest that choice of parenting strategies is influenced by internalised explanations that parents have for their child's behaviours. The authors conclude that clinical interventions can help parents alter unhelpful cognitions to enable the development of alternative explanations for their child's difficulties (e.g. challenging behaviours). This review however focused on studies conducted with children suffering from cognitive delays and not from ASD symptoms ASD, which may specifically influence the quality of parent-child interactions (e.g. McConachie & Diggle, 2007).

The association between health cognitions and subsequent behaviours can be understood using models of health-related behaviour, including 'The Health Belief Model' (Becker, 1974); 'Social Learning Theory' (Rotter, 1966) and 'Personal Construct Theory' (Kelly, 1955). The latter has been used to postulate that parents' reactions upon learning of a diagnosis are partly informed by previous expectations held by the parent for themselves and their child (Cunningham & Davis, 1985); personal expectations for parenthood are typically formed prior to a child's birth (see Dale, 1996). Accordingly, the effectiveness of diagnostic assessments or interventions will be influenced by parents' pre-existing beliefs and knowledge base formed through their own attempts to understand their child. Adaptations to pre-existing beliefs are necessary to enable parents to learn and apply new information to their child (Tucket, Boulton, Olson, & Williams, 1985; Ley, 1989). Changes to parental cognitions are associated with the quality of the parent-child relationship, as suggested by the interactions observed between depressed mothers and their children (Bolton et al., 2003).

Parent held cognitions, including appraisals of disability and its impact on the child and family, are hypothesised to mediate the relationship between child related stressors (e.g. behaviours) and parenting strategies (Hastings, 2002). For example, parents' appraisals and

beliefs about disability have been shown to influence their adaptation to the challenges of parenting a child with a disability (Trute, Hiebert-Murphy and Levine, 2007). Sameroff and Fiese (2000) propose a theoretical, cross-lagged model to outline the influence that parental cognitions may have upon child outcomes. Within this model the authors use an example of birth complications to suggest that complex interplay between external factors (e.g. disability), parent cognitions (e.g. anxiety) and subsequent behavioural reactions from both parents and children (e.g. avoidance and challenging behaviours, respectively), may augur towards certain child-related outcomes (e.g. language delay).

Following a child's diagnosis, parents have been shown to cope by seeking out disorder-specific information in order to adapt any pre-existing appraisals of disability (Starke & Mollers, 2002). Newberger's model (1980) suggests that the development of this knowledge base reflects parental understanding pertaining to the second out of the four levels. Parents of children with complex symptom presentations, such as those seen in neurodisability services, may need clinical input in order to further enhance their levels of understanding and foster better outcomes for their child.

In a non-clinical sample of 68 parents, cross-sectional questionnaire data showed an association between parental knowledge of effective parenting strategies and children's level of disruptive behaviour. This relationship was moderated by the level of parental dysfunction, such that knowledge was only related to child behaviour when dysfunction levels were low (Morawska, Winter, & Sanders, 2009). Parental dysfunction was measured using a composite score across three parenting constructs: permissive discipline, over-reactivity and verbosity. These constructs may not fully measure aspects of parenting that relate to child behaviour (e.g. parents' level of expectations and their reflective functioning abilities: Slade, 2005). These results suggest that parental knowledge of parenting strategies neither fully explains child behaviour, nor directly affects parents' abilities to understand and prepare for problems

specific to their child, in the context of other difficulties (e.g. parental dysfunction). Subsequently, for children with atypical development, disorder specific knowledge may not directly transfer to adaptive parenting strategies. Due to the cross-sectional study design, no inference can be made regarding the direction of this relationship; further longitudinal research is therefore needed.

There is some evidence within the medical literature to suggest that patient's understanding of their illness is related to disorder-specific knowledge; for example with regards to patients' knowledge of their own medical symptoms (Heisler, Piette, Spencer, Kieffer, & Vijan, 2005). In a cross-sectional survey of 686 American adults diagnosed with Type 2 Diabetes, knowledge of a specific health marker (HbA) was associated with greater accuracy in assessing glycaemic control levels, and a greater understanding of diabetes self-care. Any generalisations taken from this study with regards to parental understanding of children with ASD must be taken with caution. This study examined adults' understanding of their own difficulties and did not assess the affect that clinical input has on the relationship between knowledge and understanding. Further, similarly to previously mentioned studies which investigated parental understanding (e.g. Tunali & Power, 2002), illness understanding was assessed using a single question (*'how well do you understand how to manage your diabetes'*), which prevents assessment of construct validity or internal reliability. Whilst this study indicates a potential relationship between knowledge and understanding, the only known study to examine understanding in parents of children diagnosed with ASD, showed no relationship with parents' disorder-specific knowledge (Moran et al., submitted).

The above literature strongly argues for the importance of examining parental understanding as an outcome indicator within child neurodisability services. Parents of children with special health-care needs have been shown to prefer individualised service provision for their child (McConachie, 1994; Case, 2001). In order to offer this, services may need to utilise and



enable parental understanding, for example by tailoring information and recommendations given to parents regarding their child's specific needs, in addition to delivering disorder-specific knowledge. As noted by Glaun, Cole, and Reddihough (1998) and supported by the current literature review, few studies have directly investigated parental understanding or the impact that clinical interventions may have upon such understanding.

The PUN-Q (Moran et al., submitted) is the only identified tool which currently exists to allow systematic examination of parental understanding within the context of neurodisability (see section 1.11 below for more information). The PUN-Q however has not yet been validated for its reliability and sensitivity as a pre-post assessment outcome measure. Both the initial development and current study have focused upon validating the PUN-Q for parents of children with ASD symptoms. In order to delineate any specific effects of ASD on parental understanding, the following section will define ASD and outline the process that parents may experience when attempting to gain a diagnosis for their child.

### **1.3 Definition of Autism Spectrum Disorders**

ASD is a neurodevelopmental condition usually diagnosed in early childhood, which affects social communication abilities (Hughes, 2008). The prevalence of ASD has increased over the past four decades (see Baron-Cohen et al., 2009; Schultz, Schmidt, and Stichter, 2011). Recent studies have estimated the prevalence of Autism to be between 94 and 157 per 10,000 children (see Baron-Cohen et al., 2009).

The Diagnostic Statistical Manual – Version Four (DSM-IV: APA, 1994), stipulates that in order for a diagnosis of Autism to be reached, at least six symptoms are required; at least two showing qualitative impairments in social interaction, one or more regarding impairments in communication, and one or more regarding repetitive or stereotyped patterns of behaviour,

interests, and activities. Symptoms need to occur before the age of three in at least one of the following areas: social interaction, language and symbolic or imaginative play (APA, 1994).

As a consequence of criticisms, the latest edition: DSM-V (APA, 2013) has created the dimension of ASD, which amongst other changes, reduces ASD symptoms into two domains: social communication and fixated interests, repetitive behaviours or activities, in addition to giving more flexibility to the age criterion.

For the purposes of the current study, the term ASD will be used to encapsulate all types of social communication disorders, which are recognised either within the DSM-IV or DSM-V.

#### **1.4 Neurodisability and Neurodevelopmental Disorders**

The estimated cost for supporting families and children with ASD in the UK is approximately 2.7 billion pounds per annum (Knapp, Romeo, & Beecham, 2009). Over an average lifetime, the estimated cost per individual is thought to total 1.23 million pounds (Knapp et al., 2009). It is therefore essential that clinical input is tailored to the families' needs; appropriate outcome measures are needed in order to evaluate services' effectiveness to ensure greatest economic and health efficiency.

The diagnostic process and paediatric management of ASD is included within the broader category of paediatric neurodisability, which is a sub-speciality of Paediatrics. Health services for paediatric disability are based on a tiered model, with referrals transferred from primary to secondary care services (Tier-Two), depending on symptom severity (Department of Health and Social Security, 1976; 1978). Concerns regarding complex or rare ASD disorders are referred onto regional specialist centres (Tiers Three and Four). Such cases include those

which are borderline with unclear diagnosis, present with an atypical form, or are comorbid with another syndrome or disorder,

The National Institute for Clinical Excellence guidelines (NICE, 2011) recommend that ASD diagnostic assessments be conducted by a Multi-Disciplinary Team (MDT) consisting of health-care professionals including Psychologists, Occupational Therapists, and Speech and Language Therapists. Children with complex needs are assessed within specialist Tier-Four services. These assessments aim to integrate together information from other professionals, the parental developmental history interview, clinical assessment, and observations of the child in more than one setting (e.g. clinic, home and school) (Bruey, 2004).

Such assessments are conducted in order to develop a diagnostic opinion, provide a second opinion following a local assessment, or to help inform parents about the disorder and how it may individually affect their child (Dale & Godsman, 2000). Following the completion of an assessment report, Tier-Four services share their understanding of the child's difficulties and their treatment recommendations with different members of the wider system (e.g. carers, school and the local health-care teams). A short-term longitudinal study which compared MDT assessments to those conducted by single practitioners showed that MDT assessments significantly enhanced parents' understanding of their child's difficulties in comparison to assessments conducted by single practitioners. This study was conducted with parents of 66 children presenting with symptoms of Learning Disability or challenging behaviours, therefore it is unknown how far these results can be generalised for children with suspected ASD. Further, analyses were based upon single-item questions (e.g. *'the assessment helped us to understand our child's behaviour better'*), which restricts the findings' reliability and validity (Mittal et al., 2014).

## **1.5 The Diagnostic Process**

The Tier-Four MDT assessment which will be investigated within this thesis, aims to provide diagnostic clarification for parents and local professionals. By the time that children are referred to such services they may have experienced different diagnostic tests and assessments, and have been seen by a number of services and health-care professionals (Graungaard & Skov, 2007). Regardless of any previous clinical input, all local consultant paediatricians and parents newly referred to Tier-Four clinics retain unanswered questions regarding a child's difficulties, which the service deem worthy of further assessment. The diagnostic process, i.e. the process by which parents receive an explanation for their child's symptomatology, is often a time of much uncertainty and stress for families (Mansell & Morris, 2004). During this process parents are perceived as vulnerable with regards to their own self-perception and understanding of their child (Dale, 1996). ASD Symptoms are detectable in children from twelve to eighteen months old (Baghdadli, Picot, Pascal, Pry, & Aussilloux, 2003), and can be diagnosed from thirty months old (Gillberg, Nordin, & Ehlers, 1996). However, the estimated average age in the UK for an ASD diagnosis is four to five years old (Baird et al., 2006). Tier-Four services accept assessment referrals for children up to eighteen years old (Moran et al., submitted); this may reflect greater symptom complexity, however it is also in line with the more flexible age criterion included within DSM-V (APA, 2013).

The longer length of the ASD diagnostic process increases the time that parents experience uncertainty, lack of validation regarding their concerns, or misdiagnoses for their child (Howlin & Asgharian, 1999; Mansell & Morris, 2004). This delay can prevent parents from adapting effectively to parenting a child with special health-care needs (e.g. Cunningham & Sloper, 1977; Blacher, 1984). Earlier diagnoses are associated with reduced adverse impact on family life (Cottrell & Summers, 1990), greater perceived collaboration with health-care professionals, and lower levels of parental stress (Moh & Magiati, 2012). Siklos and Kerns

(2007) used self-report questionnaires to retrospectively investigate the experiences of fifty-six parents of children (aged between two and eighteen years old) with ASD. Families had appointments with an average of 4.5 professionals and waited approximately three years before receiving a diagnosis.

Qualitative research conducted with thirty parents of children diagnosed with 'life-limiting' disorders (e.g. severe Cerebral Palsy) (Davies, Davis, & Sibert, 2003) concluded that diagnostic confirmation is an important validation for parents' concerns, helping them to feel understood, listened to and empowered to plan for the future. These results were supported in a further qualitative study conducted with thirty-nine parents of children diagnosed with developmental disabilities (Hiebert-Murphy, Trute, & Wright, 2011).

The diagnostic period both prior to, and inclusive of an attendance within Tier-Four services, is important contextually due to its potential impact on parental understanding; parents seen within Tier-Four services may have experienced longer delays before receiving a confirmed diagnosis for their child. Graungaard and Skov (2007) conducted a qualitative study of eight couples who had children (aged up to twenty-seven months old) with physical or neurodevelopmental disabilities. Interviews were conducted three months post diagnosis and repeated after two years. Negative parental experiences during the diagnostic period were associated with parents utilising fewer constructive coping strategies. Whilst this study offers important insights into parents' diagnostic experiences, it fails to take account of potential differences between parents of children with physical or neurodevelopmental disabilities. The potentially different stressors experienced between these groups may have influenced the study's results. The young age of the children included within this study may not allow these results to be generalised to specialist Tier-Four services (which have an older average age) (e.g. Moran et al., submitted).

Child symptom complexities in addition to the waiting period for specialist services, may affect the way that parents perceive their child, their child's symptoms, and their understanding regarding the aetiology of these difficulties (Mercer, Creighton, Holden, & Lewis, 2006). It is therefore important for specialised services to address parents' pre-existing beliefs in order to enhance parents' understanding of their child and the effects of their symptoms, and to help them to build realistic expectations for future prognoses. The next section will review parents' reactions to clinical interventions in order to better appreciate the effect that the diagnostic process may have on parents.

### **1.6 The Reaction of Parents to Clinical Intervention/ Assessment**

Parental understanding can potentially help to determine a parents' role within their child's healthcare provision. Effective interventions should therefore recognise parents as the advocates for change in their children (Ho et al., 1994) and target appropriate cognitions and levels of understanding. Clinical approaches such as the 'parents as partners' model (Dale, 1996; Squires, Nickel & Eisert, 1996), or 'Family Centred Care' (e.g. American Academy of Pediatrics, 2003), perceive families as essential within the assessment and throughout any decision-making processes regarding treatment (Rosenbaum, King, Law, King, & Evans, 1998). Such approaches follow the principles of self-determination, empowerment and self-efficacy for parents (Law et al., 2003).

These approaches encourage parental involvement and help to ensure that parents can comprehend and utilise health-care information. They have been associated with increased parental adherence to treatment recommendations (Graungaard & Skov, 2007), in addition to improved well-being and resilience outcomes for children and parents, in aspects such as children's behaviour, positive parenting strategies and parental well-being (e.g. MacKean, Thurston, & Scott, 2005; Dunst, Trivette, Davis, & Cornwell, 2006).

Effective child neurodisability interventions will tailor information around a child's unique needs in order to enhance parents' understanding. This approach aims to indirectly improve child-related outcomes by ensuring parental awareness, agreement and participation in their child's treatment (Simeonsson, Edmondson, Smith Carnahan, & Bucy, 1995), thus enabling parents to both comprehend and remember information relayed to them (Ley & Spelman, 1967; Ley, Goldman, Bradshaw, Kinsey, & Walker, 1972). Improved adherence to therapy is especially important for children with complex ASD symptoms, as such disorders can require intensive and long-term treatments that may be delivered mainly within the home environment (e.g. Applied Behaviour Analysis for the treatment of Autism: Lovaas, 1993).

McConachie and Diggle (2007) systematically reviewed outcomes from 12 randomised control trials that investigated training programs for parents of children with ASD (aged one to six years old). Training methodologies varied and were implemented either at home, within a clinical setting, or both. Programs consisted of methods including psycho-education, behaviour modification and teaching parents to recognise their child's cues. The training programmes were implicated in increased maternal knowledge of autism (Jocelyn, Casiro, Beattie, Bow, & Kneisz, 1998), reduced maternal depression (Bristol et al. 1993), and improvement to the quality of parent-child interactions (Aldred, Green, & Adams, 2004). The majority of these studies were however methodologically limited due to small sample sizes and a lack of longer-term follow-up assessments. Further, the age of the children included within these studies was younger than the average for Tier-Four neurodisability services, it is therefore unclear how these results would generalise. A recent review of 30 published studies showed that only 33 percent of interventions are targeted towards parents with children older than six years. Interestingly, only two of these studies reported that the training program increased parents' knowledge base regarding their child's disability (Schultz et al., 2011);

teaching parents skills specific to their child was more effective than the provision of generalised information (Kaminski, Valle, Filene, & Boyle, 2008).

Parents are regarded as the ‘lived experts’ of their children (e.g. Goldfarb et al., 2010). However, the uneven developmental profiles, and range of complex behaviours associated with ASD (Stone & Rosenbaum, 1988), make parents more likely than professionals to over-estimate their child’s cognitive, developmental or emotional abilities (e.g. Szatmari, Simms, Ainsworth, & Hill, 1994; Gray, 1995). Such differences in opinion may reflect lower levels of understanding and can affect the parent-professional relationship (Graungaard & Skov, 2007). Diagnostic assessments have the capabilities to enhance parental understanding (e.g. Mittal et al., 2014). Evaluation regarding the quality of parental understanding at the start of the clinical process will enable services to evaluate progress, thereby anticipating any potential differences in opinion, which could affect parental adherence to treatment recommendations.

Geiger, Smith and Creaghead (2002) investigated the extent to which parental understanding of their child’s cognitive functioning matched with cognitive assessment results. Children were aged between 2.5 to 10 years old and met DSM-IV criteria for Autism. Parents over-estimated their child’s cognitive functioning pre-assessment in comparison to post-assessment results. Higher severity of cognitive impairment was associated with more disparities, with parents more likely to over-estimate their child’s abilities; conversely greater parent-professional agreement was shown for parents of children with higher IQ levels. The results from this study suggest that prior to clinical intervention parents may not fully understand their child’s difficulties or abilities.

Geiger and colleagues’ (2002) study provides cross-sectional evidence to support the call for neurodisability services to monitor levels of parental understanding. Whilst this study



demonstrates a disparity between parental estimates of their child's cognitive level and their actual abilities, it does not examine whether services are able to improve parental understanding; i.e. parents' perceptions of their child's profile. Further research is therefore needed to assess this potential outcome of clinical intervention.

A prospective longitudinal examination into the effect of a multi-disciplinary assessment was conducted with mothers of children suffering from complex developmental difficulties (e.g. Intellectual Disability, Developmental Language Disorder or Autistic Disorder). Mothers (n=40) were surveyed prior to the assessment, immediately after receiving the MDT feedback and six months post-assessment. Mothers were shown to under-estimate the extent of their child's delay, both pre-and-post assessment. Maternal estimations were more in line with professional opinions six months post-assessment, indicating a delayed increase to levels of parental knowledge about their child's development (Glaun et al., 1998). This study highlights the importance of longitudinally examining the affect of MDT assessments on parental cognitions and understanding.

The assessment and diagnostic process may act as a catalyst for changes to the quality of parents' understanding (Human & Teglassi, 1993), thereby helping parents to ascertain realistic goals for their child's future and adapt their parenting strategies. Decreased levels of parental understanding regarding a child's abilities and support needs may lead to misattributions of children's behavioural responses, or an over-estimation of their abilities; such cognitions have been linked to feelings of failure for both parents and children (Stone & Rosenbaum, 1988). For example, misattributions (e.g. perceiving a child's symptoms to be signs of behavioural non-compliance) have been linked to exacerbation of child behavioural problems, parental frustration, increased parenting stress levels, and the utilisation of harsher discipline strategies (Glascoe, 1994; Chavira, Shapiro, Blacher, & Lopez, 2000; Lecavalier, Leone, & Wiltz, 2006).

Conversely, parents whose expectations of their child's potential are too low, may provide insufficient stimulation or challenge for their children, which is also likely to negatively affect outcomes (see Rogers et al., 1992). Increased parental understanding into the expected impact of their child's neurodisability may enable parents to develop realistic developmental expectations for their child. This aim should be integral to and monitored within the clinical assessment process. The PUN-Q has been designed and initially validated as an instrument to focus upon this specific parent-related outcome (Moran et al., submitted). Further research is however needed to investigate whether this tool can effectively measure change encouraged by clinical input. Parental understanding within the context of neurodisability may also be an important indicator of other parent related outcomes, such as perceived parenting stress and parenting self-efficacy beliefs. These potential associations will be outlined within the following two sections of this chapter.

### **1.7 Factors Affecting Parental Understanding: Parenting Stress**

In addition to facilitating parents setting appropriate expectations for their child, increased parental understanding may help to determine parents' consequent coping capabilities. Models of stress and coping emphasise the role of an individual's cognitions on determining their appraisals and emotional responses to stressful situations (e.g. Lazarus & Folkman, 1984). Appreciating the extent of parenting stress is especially pertinent for parents of children with ASD, as they experience heightened stress levels in comparison to parents of either typically developing children, or children with other neurodevelopmental disorders (e.g. Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001; Smith, Oliver, & Innocenti, 2001; Boyd, 2002; Mancil, Boyd, & Bedesem, 2009). Heightened stress levels contribute towards parents misattributing challenging behaviours, having difficulties setting realistic expectations

for their child and their own parenting, and perceiving greater severity for their child's ASD symptoms (Hastings & Johnson, 2001).

Parenting stress levels have been empirically associated with the severity of child behavioural problems in both cross-sectional and longitudinal studies. For example, a cross-sectional survey of sixty mothers of children diagnosed with Pervasive Developmental Difficulties (PDD) (aged two to seven years old) showed that higher parenting stress levels were associated with child behaviour difficulties, including increased irritability, social withdrawal, non-compliance and/or a decreased ability to initiate self-care behaviours (e.g. feeding, washing, and dressing) (Tomanik, Harris, & Hawkins, 2004).

Whilst no causality inferences are possible from Tomanik and colleagues' study (2004), these results have been supported by a short-term longitudinal study which focused on the association between parenting stress and child behavioural difficulties in a sample of younger children diagnosed with PDD (aged between twenty to fifty-one months old) (Herring et al., 2006). Parents of 123 children completed questionnaires prior to and twelve months following a diagnostic assessment. Child behavioural and emotional problems were significantly associated with poorer parental mental health and greater perceived parenting stress; these relationships retained stability over time (Herring et al., 2006). However, this study was conducted with young children and parenting stress has been shown to increase as children get older (Shearn & Todd, 1997; Tonge & Einfeld, 2003). Consequently, further longitudinal research is needed on a sample of older children before any firm causative conclusions can be made.

It is unclear within the literature whether the severity of children's symptoms alone can explain parenting stress levels (see review by Hassall & Rose, 2005). Not all parents of children with disabilities experience prolonged distress (Benzies et al., 2011); the majority of

parents show effective adaptation to their role as caregivers (Hassall & Rose, 2005). Subsequently, in order for assessment services to provide appropriate help for families, it is necessary to ascertain factors which promote or preclude successful adaptation. The review by Hassall and Rose (2005) concludes that stress cannot fully explain parental coping difficulties. A more complex model involving parental cognitions of disability is instead implicated. Subsequently, systematically investigating differences in parents' understanding with a measure such as the PUN-Q may allow services to gain greater insight into parental resilience, for example, whether parents with higher levels of understanding are better able to cope with the demands of parenting a child with ASD.

High levels of coping are necessary for the long-term commitment and responsibility required in parenting a child with a neurodisability (e.g. attending frequent appointments with different health-care professionals) (see Dumas et al., 1991). In addition to the time commitments, children with ASD exhibit greater behavioural and emotional difficulties, in comparison to children with other neurodevelopmental disorders or cognitive delay (Tonge & Einfeld, 2003). These children are less able to communicate or respond appropriately, therefore placing extra strain upon parent-child interactions (Johnson & Myers, 2007). These behavioural, emotional and communication difficulties can persist over time (Baker, Blacher, Crnic, & Edelbrock, 2002), contributing towards increased stress and poorer mental health outcomes for parents (Beck, Hastings, Daley, & Stevenson, 2004); for example, depression (Chilcoat & Breslau, 1997) and symptoms of post-traumatic stress (Baylot-Casey et al., 2012).

It is important to identify the levels of stress experienced by parents, as left untreated, high stress levels are associated with parents utilising fewer coping resources. For example, parents may be less likely to bring their child to health-care services, which would diminish the quality of treatment received (Mowery, 2011).

In addition to influencing parent's ability to adapt, parents' cognitions (i.e. emotional reactions and appraisals) regarding parenting a child with a disability will influence the level of parenting stress experienced (Hastings, 2002) and determine the effect of the stress upon parenting strategies and subsequent child-related outcomes (Webster-Stratton, 1990). In a theoretical model which links parenting stress and child behaviour problems, parental cognitions (which form part of parental understanding) have been proposed to mediate the relationship between the utilisation of less effective parenting strategies (e.g. coercive parenting) and increased child behavioural problems (Hastings, 2002). This model hypothesises a role for specific cognitions in contributing towards parenting stress. It is therefore necessary to add to this model by exploring the association between parental understanding and parenting stress.

Moran and colleagues (submitted) directly examined the association between parental understanding of their child's neurodisability and parenting stress, in order to assess construct validity for the new PUN-Q scale. Fifty-nine parents of children diagnosed with ASD were surveyed following the completion of a Tier-Four diagnostic assessment. Results suggested that higher PUN-Q total scores (i.e. parental understanding) were associated with lower levels of parenting stress (Moran et al., submitted). Further research is needed to assess the prospective relationship between parental understanding and parenting stress and how this may change over time.

In order to examine this relationship, Moran and colleagues (submitted) utilised the Parenting Stress Index – Short Form (PSI-SF; Abidin, 1995). The PSI-SF is a standardised and widely used self-report questionnaire, which has been used to measure stress for parents of children experiencing symptoms of ASD and developmental delay (e.g. Hassall, Rose, & McDonald, 2005; Davis & Carter, 2008). The PSI-SF measures parenting stress across three domains:

parental distress, parental-child dysfunctional interactions, and parental perceptions of how difficult their child is to manage. This is a useful instrument to utilise with the PUN-Q, in order to be able to specify the domains of stress which are most likely to be associated with parental understanding. Such information would help services to tailor their clinical provision in order to decrease parenting stress and therefore reduce the likelihood of negative child or parent related outcomes.

The studies identified within this short review suggest that an association exists between increased parenting stress and both parental and child related outcomes (e.g. Glascoe, 1994; Chavira et al., 2000; Hastings, 2002; Mowery, 2011). These outcomes may potentially influence or be influenced by parental understanding (e.g. Glaun et al., 1998). Further research is needed to assess these relationships over time in order to help clinical services better understand and target parental risk and resilience factors.

### **1.8 Factors Affecting Parental Understanding: Perceived Parental Self-Efficacy**

Parenting a child with ASD is the equivalent of experiencing a long-term and unpredictable stressor (Norton & Drew, 1994). In order to feel able to effectively parent a child with disabilities, parents must first understand their child's ongoing and changing needs. Subsequently, parenting stress, which has been shown to affect how parents are able to understand and adapt to their child's behaviour (e.g. Chavira et al., 2000; Hinshaw, 2002; Sameroff & Fiese, 2000), may also affect their beliefs regarding their parenting competence. Taking into account the potential association between parental understanding of a child's neurodisability and parenting stress, parental understanding may also affect, or be related to, parents' levels of perceived self-competence (e.g. Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006).

Perceived parental self-competence is also referred to as self-efficacy (Bugental & Johnston, 2000), or parenting self-esteem (Johnston & Mash, 1989); these terms are used interchangeably within the literature (see Hassall & Rose, 2005). Parental self-efficacy beliefs infer how effective parents perceive themselves to be within their care-giving role (Hassall et al., 2005; Jones & Prinz, 2005), within the context of neurodisability. This may relate to how confident parents feel in coping with their child's developmental difficulties (Ardelt & Eccles, 2001). In their review of parental self-efficacy, Jones and Prinz (2005) conceptualised three separate domains of self competence: parents' general feelings of competence, their feelings of competence regarding a range of parenting tasks, and their feelings of competence with respect to specific parenting domains (e.g. discipline or communication).

In typically developing children, this understanding and subsequent parental self-efficacy is partly influenced by parents' abilities to utilise 'Reflective Functioning'. 'Reflective Functioning' refers to parents' abilities to understand the factors influencing their child's behaviours and emotional states (Fonagy, Steele, Steele, Moran, & Higgitt, 1991). ASD challenges parents' Reflective Functioning due to its noted impact on social communication and interaction abilities (e.g. APA, 1994). It is possible that parents of children with ASD may find it harder to understand or reflect about their child's atypical and unpredictable social responses (van Ijzendoorn et al., 2007); this will make it harder for such parents to achieve higher levels of perceived self-efficacy.

Factors that may influence parental self-efficacy, such as parental understanding, are important to investigate within child health-care settings due to the association between perceived parenting self-efficacy and children's behavioural and developmental outcomes (see Jones & Prinz, 2005). For children with special health-care needs, parenting self-efficacy ratings have been shown to influence the extent to which parents feel competent to meet their

child's higher complexity of needs (Teti & Gelfand, 1991) and to comply with healthcare recommendations (Calvert & Johnston, 1990). In a review by Giallo, Kienhuis, Treyvaud, and Matthews (2008), higher parental self-efficacy was related to increased use of positive parenting strategies and persistence in demanding parenting situations.

Understanding the mechanisms which influence parenting self-efficacy will help services target any vulnerability with regards to this self concept. This is important as parenting self-efficacy is related to better child and parental outcomes. For example, typically developing children of parents with higher self-efficacy beliefs exhibit greater levels of enthusiasm, compliance and affection (Coleman & Karraker, 2003). This may be due to parents modelling positive attitudes, beliefs and behaviours to their child (Ardelt & Eccles, 2001). The children are consequently more likely to develop stronger self-efficacy beliefs for themselves and be more willing to challenge themselves to enhance their developmental progress (Bandura, 1997).

Whilst there are a paucity of studies directly investigating the association between perceived parenting self-efficacy and parental understanding within the context of child neurodisability, results from other fields suggest that there may be a positive association. For example, self-competence and self-rated understanding has been correlated in areas such as diabetes (Heisler et al., 2005); students' assessments of their academic abilities (Mabe & West, 1982); and people's beliefs regarding career progression (Brown, Lent, & Gore, 2000).

Parents of children with ASD who feel able to positively enhance their child's development have been shown to retain higher levels of parenting self-efficacy and lower levels of parenting stress (Hassall et al., 2005). Conversely, low levels of perceived competence have been shown to be related to greater maternal depression and parenting stress in a cross-



sectional survey of 170 mothers of children with Autism (Kuhn & Carter, 2006); no interpretation can be made however regarding the direction of these associations.

Children's developmental successes strengthen parent's beliefs in their own parenting abilities (Elder & Conger, 2000). For children with neurodisabilities, the threshold for success may need to be adapted due to the impact of the disorder. Understanding the ramifications of their child's neurodisability will help parents to set realistic expectations and appreciate any achievements from their specific and tailored parenting strategies. In a cross-sectional study 29 mothers of children with Autism were compared to 29 matched mothers of typically developing and healthy children (aged between 5 and 14 years old). The mothers of children with Autism reported greater difficulty understanding their child's behaviours, despite spending significantly more waking hours with their child; the two groups did not differ regarding the perceived importance of understanding their child (Tunali & Power, 2002).

Results from Tunali and Power's (2002) study showed that parental understanding in conjunction with parents' self-efficacy beliefs was related to life satisfaction for mothers of children with Autism. Contrastingly, perceived self-efficacy was not associated with life satisfaction ratings for mothers of typically developing children (Tunali & Power, 2002). The mothers of children with Autism also placed greater value on perceiving themselves to be 'good mothers'. This emphasis on parental responsibilities is likely to place extra pressure upon these parents within their caregiving role. This difference between mothers of children with Autism and those with typically developing children may help to explain the aforementioned negative association between parental stress and sense of competency (Hassall et al., 2005).

Tunali and Power's (2002) study indicates an important role for parental understanding with regards to maternal well-being and life satisfaction, however the reliability of the results is

limited by the hitherto lack of a standardised instrument for measuring parental understanding. This study has however cautiously highlighted the importance of parental understanding within the experience of those parenting children with ASD. These results therefore provide further support for the development of a new instrument which can systematically measure parental understanding of their child's individual neurodisability (e.g. Glaun et al., 1998; Moran et al., submitted).

It can also be deduced from this study that parental understanding is difficult to attain for parents of children with ASD; time spent together does not guarantee increased understanding (Tunali & Power, 2002). It is therefore important that interventions focus on helping parents to improve their understanding in order to enable them to gain a greater sense of self-efficacy in their role as parent and caregiver, and potentially more realistic expectations against which to measure success. Neurodevelopmental assessments could potentially help parents by providing them with a scaffold against which they can understand their child's development (see Dale, 1996). Such clinical input can help parents to anticipate future parenting demands, to acknowledge parenting successes, and realistically evaluate their role in helping their child to meet appropriate developmental goals.

Reliable and valid outcome measures are necessary in order to evaluate the effectiveness of such assessments. The Parenting Sense of Competence Scale (PSOC: Johnston & Mash, 1989) has been identified as the most commonly used instrument for measuring parenting self-efficacy (Jones & Prinz, 2005). This instrument assesses general parenting self-efficacy beliefs and is therefore appropriate for parents of children with neurodevelopmental disorders; it does not focus on specific parenting tasks that may not be relevant to parents of children with ASD. The PSOC consists of two validated subscales: *Satisfaction* (i.e. feelings regarding parenting that the parent may have experienced within their care-giving role), and *Efficacy* (i.e. the extent to which parents feel able to apply parenting strategies). This measure has

been validated for use with both mothers and fathers, and can be used by parents of older children and adolescents (Johnston & Mash, 1989), unlike other self-efficacy instruments which are age dependent (e.g. the Toddler Care Questionnaire: Gross & Rocissano, 1988). This is important with regards to ASD, in which complex cases may have to wait longer before diagnostic confirmation (Dover & LeCouteur, 2007).

The literature has thus far outlined the potential effects of diagnostic assessments for both parents and children. The following section will review different ways in which effectiveness is currently being monitored.

### **1.9 Assessing Service Efficacy**

The quality of the neurodevelopmental assessments is therefore essential to help enhance parent-related outcomes, such as perceived parental understanding, parenting self-efficacy beliefs and reductions to parenting stress. In order to ensure that these aims are met, assessments must be continuously monitored (e.g. Department of Health, 2005; Office of Health Economics, 2008). By necessity, part of the clinical governance process requires service providers to seek out appropriate measurement tools which are sensitive and specific to different aspects of health-care provision. The challenge for health-care providers is to identify and utilise the measurement tools which tap into the most important aspects of patient care.

Effectiveness of clinical interventions can be measured by patients' functional improvement, or by their perceptions regarding changes to their Quality of Life (QoL) (see Fayed et al., 2012). A further construct labelled 'Health-Related Quality of Life' relates to a patient's *perceptions* about their health (e.g. their personal goals, expectations, and satisfaction with regards to their levels of functioning) (see Fayed et al., 2012).

The mostly widely used Patient Reported Outcome Measures (PROMs) within clinical empirical studies are those which have proven psychometric properties (including standardisation and validation). Fayed and colleagues (2012) conducted a systematic review of all articles published between January 2004 and December 2008, in order to assess the appropriateness of the utilised measure with regards both to the aims of the study and its intended measurement purpose. The results showed an inconsistency between the stated aims of the study and the specific purpose of the outcome measure utilised. Specifically, there was an over-reliance on measurements of functional health to assess aspects of HRQoL; only four PROMs of the fifteen identified were coded by the reviewing team to focus intentionally on HRQoL (according to the WHO definitions) (Fayed et al., 2012).

Fayed and colleagues (2012) conclude that researchers are utilising well established PROMs whilst compromising on their abilities to measure the intended construct, which limits the reliability of results (Fayed et al., 2012). This review highlighted a paucity of published HRQoL PROMs, or Patient Reported Experience Measures within the literature. A further limitation, which was also noted within the review, is the over-reliance on cross-sectional studies (Fayed et al., 2012). This is an issue which may be particularly relevant to specialist Tier-Four services (including neurodisability), due to the relatively short time scale that such services are involved with families (e.g. one-off, short-term multiple assessment episodes, bi-annual, or annual review appointments). Such short-term or sporadic involvement within clinical services augurs towards cross-sectional, rather than longitudinal or causative research designs.

Two measures which are widely used within children health-care services due to their ease of use, strong validation, and focus on the complexity of child functioning, are the 'Strengths and Difficulties Questionnaire' (SDQ: Goodman, 1997) and the 'Social Communication

Questionnaire' (SCQ: Rutter, Bailey, Berument, Lord, & Pickles, 2003). These measures can be completed by parents or teachers on behalf of children, or in the case of the SDQ, by self-report. The SDQ assesses well-being across five constructs: emotional symptoms, conduct problems, hyperactivity/inattention, peer-problems and pro-social behaviour. Diagnosing ASD requires a mixture of clinical interviews, structured assessments (e.g. the Autism Diagnostic Observation Schedule: Lord et al., 1989) and observations. This process is lengthy and costly. The SCQ therefore provides clinicians with a brief overview of a child's potential risk for social communication difficulties, by asking parents about characteristic symptoms of ASD. It is used by clinicians as a brief and reliable screening questionnaire for ASD (Berument et al., 1999)

### **1.10 Measuring Assessment Effects on Parents**

In addition to directly affecting child-related outcomes, any effective neurodisability intervention or assessment must take levels and quality of parental understanding and cognitions (including concerns and expectations) into consideration. Limited input and resources from specialist services places much of the care-giving burden onto families (see Dale, 1996). A challenge faced by neurodisability assessments is therefore professionals' abilities to effectively communicate with parents so that relayed information can be understood, retained and utilised (Ley, 1989). Accordingly, services must find ways to accurately measure parental experiences of their child's assessment, using short and appropriate measures; tapping into both functional health-related and QoL constructs.

The most widely used outcome measure which focuses on parental perceptions within child-disability is the 'Measures of Processes of Care' (MPOC: Rosenbaum, King, & Cadman, 1992). This 56 item instrument (revised to 20 items: King, King, & Rosenbaum, 2004) measures parents self-reported perceptions regarding the quality of care provided by

professionals: for example, *'to what extent do the people/ centre who work with your child...'*. The MPOC has been utilised and validated for children with a wide range of neurodisabilities (Larsson, 2000; McConachie & Logan, 2003). Whilst it provides useful information for improvement to services that provide ongoing health-care provision, it is less useful for specialist neurodisability services which provide short-term, comprehensive and sporadic diagnostic assessments. Further, focusing on the provision of care by professionals does not allow measurement of the effect of a service on parents' understanding of their child, or their perceptions of their own efficacy as carers.

Alternative scales used within the literature also focus mainly on auditing parents' perceptions of health-care provision. For example, the 'Family Focused Intervention Scale' (Mahoney, O'Sullivan, & Dennebaum, 1990) includes 40 items assessing parental perceptions of healthcare across five domains including child information; personal family assistance and resource assistance. Similarly, the 'Family Centred Program Rating Scale' (Murphy, Lee, Turnbull, & Turbiville, 1995) audits the provision of family-centred healthcare; this scale is aimed at early intervention and therefore is not appropriate for assessment based services or long-term health-care provision.

The 'Parenting Morale Index' (Trute and Hiebert-Murphy, 2005) focuses on parents' cognitions directly related to parenting a child with a disability include. This measure consists of 10 items rated on a five point Likert scale to examine the extent to which parents feel positive within their role as caregivers. The 'Family Impact of Childhood Disability' scale (Trute et al., 2007) consists of 20 items to measure parents appraisals of the impact upon the family of having a child with a disability. Due to their focus on impact, these measures are useful in identifying parents with increased psychological risk, however they do not measure parental understanding, which is potentially a separate construct that could also influence parents' psychological factors.

Further outcome measures exist (e.g. 'The Diagnostic Survey': Howlin & Moore, 1997), however these also seem to focus mainly on the parent-professional relationship, in addition to the quality, availability and accessibility of services. Whilst these aspects of health-care provision are undeniably important, other previously argued factors (i.e. parental understanding, parenting stress and parenting self-efficacy) are theorised to impact on parental utilisation of services and their compliance with professionals' treatment recommendations. These factors include parental understanding of the diagnosis and treatment recommendations, in addition their perceptions of the impact on their child of any symptoms associated with neurodisability diagnoses (see Glaun et al., 1998). Previous studies have indirectly attempted to examine the effect of the diagnostic assessment upon parental understanding by comparing parents' pre-assessment perceptions to professional assessment findings (e.g. Geiger et al., 2002; Ho et al., 1994). Whilst these studies infer an underlying construct of parental understanding, they do not measure it scientifically nor recognise it as a potentially independent construct which requires a separate validated instrument.

Alternative methods have utilised qualitative interviewing (e.g. Roden, 2003), which can be overly time consuming and therefore not ecologically viable within the demands of a clinical assessment service, nor suitable for within-population comparisons. Furthermore these interviews do not provide a systematic measure which lends itself towards longitudinal research designs. For example, in postulating the aforementioned construct of 'Parental Awareness', Newberger (1980) formulated a semi-structured interview which aimed to examine the different factors related to parents' thoughts and behaviours in their parenting role. This interview is lengthy and targeted towards parents of typically developing children; it therefore includes many questions not relevant for parents of children with disabilities (e.g. regarding parent-child conflict).

Authors have called for the examination of parental understanding due to the effect that it may have for children with special health-care needs (e.g. Simeonsson et al., 1995; Glaun et al., 1998). Whilst some studies have unsystematically or indirectly examined the relationship between parental understanding and child or parent outcomes (e.g. Tunali & Power, 2002), none of the identified published studies have utilised an instrument which specifically measures parental understanding within the context of child neurodisability and its individual effects on their child; studies have instead incorporated one or two items within a larger set of questions. The PUN-Q measure (Moran et al., submitted) used within the current study is the first instrument which has been developed to systematically measure parents' understanding of their child's difficulties and the impact that these symptoms may have upon their child.

### **1.11 Developing a measure of Parental Understanding: The PUN-Q**

The PUN-Q (Moran et al., submitted) was developed and validated cross-sectionally using retrospective data collected from 59 parents of children who received a diagnosis of ASD following a Tier-Four MDT Neurodisability assessment between 2010 and 2011. The PUN-Q was developed through an iterative process of development, including qualitative interviewing of a small sample of parents to aid the item generation process (Flick, 2009; Weber, 1990), Delphi rating for content validity using an expert professional panel (Lynn, 1986), and then finally an initial validation study leading to a psychometric statistical analysis. Please see Appendix 5 for more information regarding the item generation and content analysis process. The following sections will outline the completed factor analysis, reliability and validity examination in order to identify the further investigations which are needed for this new instrument.



### 1.11.1 PUN-Q Factor Analysis.

Exploratory factor analysis was conducted to enable examination of any underlying latent factors within the thirteen included items; this is a common approach for the analysing the structure of new scales (Fabrigar, Wegener, MacCallum, & Strahan, 1999). Maximum likelihood factor analysis was used to extract the factors, to enable testing of the significance of factor loading and inter-factor correlations (Fabrigar et al., 1999). This was followed by oblique rotation of the factors, which is a process that has been shown to improve the interpretability of factors and does not assume non-independence (i.e. factors are allowed to correlate) (Field, 2009). The sample size ( $n=59$ ) did not satisfy either stringent or more lenient requirements for effective factor analysis (i.e. participant item ratios of either 10:1 or 4:1: Tabachnick & Fidell, 2007; Breakwell, Hammond, & Fife-Shaw, 2000, respectively). A recent review has however concluded that smaller samples can be used when a limited number of well-defined factors are extracted (deWinter, Dodou, & Wieringa, 2009).

Following factor analysis of PUN-Q-13, three factors were extracted with eigenvalues greater than Kaiser's criterion of 1.0 (Field, 2009). Together these factors explained 65.9% of the variance. The first factor was interpreted as representing parents' 'post-assessment understanding' of their child (e.g. *'explanations that I have been given to explain my child's difficulties make a lot of sense to me'*) and accounted for 42.9% of the variance. The second factor was interpreted as representing parents' 'insightful understanding' of their child (e.g. *'most of the time, I understand why my child behaves the way that s/he does'*) and accounted for 13.3% of the variance. The third factor was interpreted as representing parents' 'application of understanding' (e.g. *'I know how to adjust what I do as a parent to take account of my child's difficulties'*), which accounted for 9.69% of the variance.

This first factor was shown to capture most of the variance within parental responses and is arguably the strongest measure of what this thesis has referred to parental understanding.

However, all three latent factors were found to strongly correlate with each other and are therefore hypothesised to represent a unified construct, which is referred to within this thesis as parental understanding of their child's neurodisability.. Moran and colleagues' (submitted) study was conducted post-intervention. In order to be able to utilise the PUN-Q both pre-and-post assessment, a shortened pre-assessment version is necessary, as the 'post-assessment understanding' factor may contaminate or confound any data collected pre-assessment; reliability and validity for the shortened pre-assessment PUN-Q-8 has not yet been investigated.

#### **1.11.2 Internal Reliability.**

A reliable scale shows a high correlation between the value of an item measured using the scale, and the true score of the unobservable latent variable ( DeVellis, 2003). Within the first study (Moran et al., submitted), the PUN-Q showed high internal reliability (Cronbach's  $\alpha=.88$ ). A Cronbach alpha of .70 is considered adequate for new scales (DeVellis, 2003). This indicates that the items within the scale are strongly related to one-another (Cronbach's alpha: Cronbach, 1951), without violating multicollinearity assumptions; i.e. no correlations were greater than  $r=.90$  (Tabachnick & Fidell, 2007). It is important to note that internal reliability is examined in place of a direct association between the latent variable and the scale's items, which cannot be attained due to the impossibility of observing the latent variable (DeVellis, 2003). As a consequence of it being a behavioural questionnaire, the PUN-Q is therefore only able to provide a proxy for the latent (i.e. unobservable) construct of parental understanding (DeVellis, 2003).

#### **1.11.3 Construct Validity.**

Validity of a scale assesses whether or not it measures what it is intended to measure (Howitt & Cramer, 2005). In addition to content validity (see Appendix 5), validity is typically

examined using both criterion and construct validity (Cronbach, 1971). Criterion validity compares the scores for a new measure against a pre-existing standardised tool that measures the same construct (DeVellis, 2003). It was not possible within either the initial or current studies to examine this due to the absence of a pre-existing measure of parental understanding with regards to neurodisability (DeVellis, 2003). Both studies therefore relied instead on examination of construct validity (Guyaat, Walter, & Norman, 1987).

Construct validity is examined by comparing total scores of the new instrument against measures of other constructs, which are hypothesised to be related (DeVellis, 2003). With regards to the PUN-Q, this was examined by relating performance on the PUN-Q to parental ratings on two standardised instruments: the 'Parental Sense of Competence' (PSOC) and 'Parenting Stress Index – Short Form' (PSI-SF). The results from Moran and colleagues (submitted) study indicated good construct validity, with significant Pearson's correlations in the hypothesised directions for both the PSOC ( $r=.38$ ,  $p<.01$ ) and the PSI-SF ( $r=-.40$ ,  $p<.01$ ). This study assessed construct validity up to two years post-intervention (Moran et al., submitted). No study has yet examined pre-intervention construct validity for the PUN-Q.

#### **1.11.4 Test-retest reliability.**

In order to ascertain whether a scale measures the intended latent construct in a consistent manner, it is necessary to prospectively assess its' performance over two stable (i.e. non-intervention) time points within the same group of people (DeVon et al., 2007). No clinical intervention which could change the construct being examined should occur between these time points (Guyaat, Kirshner, & Jaeschke, 1992). Validation of the PUN-Q to-date has been conducted using a cross-sectional, retrospective design. Consequently, it is necessary to prospectively assess its psychometric properties over a test-retest period.

#### **1.11.5 Sensitivity to detect change.**

The PUN-Q was developed as an evaluative quantitative instrument (see Guyaat et al., 1992), which should therefore be sensitive to changing levels of parental understanding over time; if parental understanding varies within parents (see Guyaat et al., 1992). In addition to measuring reliability and validity, the PUN-Q's usefulness is also determined by its 'responsiveness' (Guyaat et al., 1987). Sensitivity or responsiveness is defined by a scale's ability to detect small but important 'in state' changes over time (Guyaat et al., 1992). In order to achieve this, Guyaat and colleagues (1987) advises that multiple pre-and-post assessment measurements are taken, which can help to estimate a scale's temporal variability. Accordingly, a second validation phase is needed which will examine whether the PUN-Q is sensitive enough to pick up changing levels of parental understanding, when assessed both prior to and following a neurodisability assessment.

#### **1.11.6 Clinically significant change.**

Significance levels with regards to changes in mean values cannot solely infer the magnitude of the effect size (Kazis et al., 1989). Further, conventional p-values may not indicate whether a change is perceived to be important by the patient (Wyrwich, Bullinger, Aaronson, Hays, Patrick, & Symonds, 2005). Within healthcare settings, qualitative minimal changes could be referred to as 'clinically significant' as long as they are purposeful or meaningful to the individual patient (Wyrwich et al., 2005). Lydick and Epstein (1993) outline two methods for assessing clinically significant changes: anchor and distribution based methods. Anchor based methods refer to observable and person specific behaviour changes. These changes could be minimal, but perceived as important by the patient. Distribution based methods examine quantitative changes between mean and standard deviation values (Lydick & Epstein, 1993). The most effective analysis of change is thought to combine both of these methods (Wyrwich et al., 2005).

Effect sizes are one quantitative method for assessing the magnitude of clinical change. Effect sizes are derived by computing the difference between pre-and-post assessment means, and dividing this change score by the sample's standard deviation; effect sizes are therefore sample dependent and dependent upon homogeneity of variance (Kazis et al., 1989). Importantly, the magnitude of post-assessment change can be affected by baseline difficulty levels; lower initial difficulties augur towards greater levels of improvement (Hays and Hadorn, 1992). Consequently, with regards to the PUN-Q, any post-assessment changes could be related to baseline PUN-Q total scores, or to the potentially associated constructs of parenting stress or self-efficacy beliefs.

### **1.12 Summary**

The above literature review demonstrates the need for a systematic instrument with which to measure parental understanding, within the context of child neurodisability. A previous study has developed the PUN-Q and has conducted initial cross-sectional reliability and validity checks (Moran et al., submitted). This review has highlighted potential associations between parental understanding and parent related factors including parenting stress and perceived self-efficacy, in addition to child-related emotional, behavioural and social communication difficulties. The limitations to the previous study and has indicated a need for further, prospective validation of the PUN-Q scale.

### **1.13 Aims and Hypotheses**

The current study aimed to prospectively and longitudinally examine whether or not the newly validated PUN-Q can be used to examine pre-and-post assessment outcomes. This study also aimed to examine whether the PUN-Q is sensitive to measuring changing levels of parental understanding over time and in response to clinical intervention (in the form of an comprehensive MDT assessment). In order to do this, parents were sampled at three different time points: twice before their child attended a Tier-Four multidisciplinary diagnostic assessment, and at one time point following the assessment episode. The design of the study focused on four different objectives:

- 1) To establish prospective Construct Validity by comparing the PUN-Q to previously identified parental-related measures, which are hypothesised to be related to parental understanding both pre-and-post assessment. This was examined using standardised measures of:
  - a. Parenting stress measured by the 'Parental Stress Index – Short Form' (Abidin, 1995).
  - b. Perceived parental self-efficacy measured by the 'Parental Sense of Competence' Scale (Johnston & Mash, 1989).
- 2) To examine the test-retest reliability of the PUN-Q (i.e. whether it is stable over time) by comparing the two pre-assessment time points.
- 3) To examine whether the PUN-Q is sensitive to changes in parental understanding over time as result of intervention, by examining pre-and-post assessment levels of parental understanding. This will provide an initial examination into whether or not a multi-disciplinary Paediatric Neurodisability assessment can improve levels of parental understanding.
- 4) To understand more about factors which may influence parental understanding, by investigating the relationship of the PUN-Q to child complexity factors:

- a. The child's emotional and behavioural difficulties measured by the 'Strengths and Difficulties Questionnaire' (Goodman, 1997).
- b. The child's social communication difficulties measured by the 'Social Communication Questionnaire' (Rutter et al., 2003).

It was hypothesised that pre-assessment measurements would show:

- 1) a positive association between the PUN-Q total score with the total score and subscales for perceived parental self-efficacy.
- 2) a negative association between the PUN-Q total score with the total score and subscales for parenting stress.

It was hypothesised that post-assessment measurements would show:

- 1) Increased levels of the PUN-Q total score.
- 2) A positive association between the PUN-Q total score and an increased total score for perceived parental self-efficacy.
- 3) A negative association between the PUN-Q total score and a decreased total score for parenting stress.

It is not known whether the PUN-Q total score is related to the child's emotional, behavioural or social communication difficulties, as measured by the SDQ or SCQ. The statistical analyses therefore had no a-priori assumptions.

## **CHAPTER 2: METHODOLOGY**

### **2.1 Overview**

This study represents the second phase of a project conducted in 2011, which previously developed and initially validated a new measure – The Parental Understanding of Neurodisability Questionnaire (PUN-Q: Moran et al., submitted manuscript).

The current study recruited an independent sample of participants, who could be prospectively followed up over time, in order to further validate the PUN-Q. The first set of aims for this study focused on assessing the PUN-Q's construct validity with other standardised parent related outcome measures, and its stability over time (i.e. test-retest reliability). Next, this study aimed to examine the PUN-Q's sensitivity to detect changing levels of parental understanding, following the administration of a multi-disciplinary Tier-Four diagnostic assessment for child neurodisability. Linked to this, the current study aimed to provide a preliminary examination of whether or not the multi-disciplinary assessment effectively increases parental understanding. Finally, this study aimed to examine the relationship between the PUN-Q measure of parental understanding and standardised measures of child emotional, behavioural and social communication difficulties.

### **2.2 Participants**

Participants were 37 parents whose child had been newly referred to a regional Tier-Four paediatric neurodisability service within a large children's hospital. All referrals were received from local consultant paediatricians (following local assessments and diagnostic investigations). This is a consecutive sample and all parents who were eligible for participation were invited to participate. Parents were recruited at the point of initial referral



to the service. A total of 74 parents were invited to take part; 50% consented (mean age of parents=43.36 years old, sd=7.93). See Table 2.1 for a full break-down of sample characteristics. The sample was comprised mainly of Caucasian, well educated mothers (n=32, 86.5%) who were either full-time carers for their child (n=12, 34.3%), or in part-time employment (n=17, 48.6%). Parents were invited to participate in this study between the months of September 2013 to May 2014.

The clinic receives referrals for children suffering from a wide range of complex and rare disorders, which can be associated with symptoms including social communication difficulties, language disorders, behavioural and motor coordination difficulties, attentional, concentration and executive functioning difficulties, in addition to Learning Disabilities. In order to increase the homogeneity of the sample, and to be comparable to the initial phase of this study (Moran et al., submitted), which was conducted within the same clinic with a separate sample of parents, only parents of children referred to the clinic for questions regarding social communication/ possible ASD were invited to participate. Homogeneity within samples is desirable in order to decrease random or non-random variability within the sample (i.e. the extent to which variability within the sample are due to differences between the children's symptoms), and thereby increase the probability that observed relationships are a consequence of the variables being investigated (Prince, 2003).

Children with queries regarding ASD symptoms were chosen as the appropriate group to investigate, as children with queries regarding social communication difficulties are the largest diagnostic group of children seen by specialist child neurodisability services (Fombonne, 2009), and were commonly referred to the assessment clinic utilised for this study, which therefore increased the recruitment sample pool.

Due to the complexity of the children's symptoms, the children included within this study were on average 9.73 years old at referral ( $sd=3.66$ ). Difficulties with regards to the length of the diagnostic process have been highlighted within Chapter 1 of this thesis (e.g. Mansell & Morris, 2004). The older age of the children diagnosed with ASD may have allowed parents time to gain a natural understanding of their child's strengths and difficulties. This potentially greater stability in parental understanding may therefore have enhanced the reliability that any increases to understanding following clinical input could be attributed to the service received. Further, as a consequence of the complexity of the children seen by this clinic, and in relation to the sensitivity of carrying out this kind of research when parents are about to embark on a stress-inducing diagnostic assessment, only parents of children aged five years or older were invited to participate within this study\*.

All new referrals to the clinic were screened by the researcher to ensure that the referral was associated with social communication difficulties/ ASD. Diagnostic status was not an exclusion criterion; children who had previously received a diagnosis of ASD were also eligible for inclusion within the study, 12 of the 33 children had received a prior neurodevelopmental diagnosis within the past two years. It is important to note that all parents were referred to this specialist clinic in order to better understand some aspect of their child's presenting symptoms; all referrals accepted to the study consisted of unanswered questions regarding social communication. Accordingly, even those parents of children with a prior diagnosis retained a level of uncertainty regarding their child's symptom presentation, which warranted further assessment. Additionally, some children were seen by the clinic for a second opinion or further information regarding a previous diagnosis. Pre-assessment diagnostic status was a factor which was taken into account within the analyses.

In summary, inclusion criteria for the study were:

- parents had children newly referred to the clinic service
- parents had questions regarding their child's diagnosis or neurodevelopmental symptom presentations
- the referred child was aged between 5 and 17 years old<sup>\*</sup>
- parents spoke English as a first language
- parents or guardians were the long term carers for the child (i.e. the child was not in foster-care)
- the child may or may not have received a previous diagnosis of a neurodevelopmental disorder including ASD

## **2.3 Setting**

The Tier-Four paediatric clinic included within this research specialises in providing multi-disciplinary assessments, expert diagnostic opinion, second opinions, and treatment guidance for the management and care of complex neurodevelopmental disorders in children aged from birth to 17 years old. The clinic aims to assess children who have been too difficult to assess by local specialist services, to provide second opinions, or to recommend appropriate treatments for local teams (Dale & Godsman, 2000).

Following NICE (2011) guidelines, the multi-disciplinary team is comprised of a consultant Paediatrician, Specialist Registrar in training, Clinical Psychologist, Occupational Therapist and Speech and Language Therapist. The composition of the team varies for each child, depending on the difficulties indicated within the referral letter. Routinely, the clinical team

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<sup>\*</sup> The minimum age criterion was not stipulated within the study's ethical approval, therefore parents with younger children were initially included within the study. The researchers decided to initiate the minimum age criterion following qualitative feedback from parents regarding the stress that they were experiencing in trying to gain a diagnosis for their child. In order to alleviate the burden, parents initially included who had children under the age of five years were only contacted by post regarding the study follow-ups and were not telephoned by the researcher.

liaises with the local team in order to ensure continuity of health-care. However, any decisions reached within the assessment procedure are made independently of the local team.

In order to be accepted for an assessment within the clinic, children must be referred by a Paediatrician. Most children seen within the clinic have therefore undergone previous assessments within their local Child Development Team, which have identified them to have a neurodevelopmental disability, without a specific diagnosis having been established or agreed upon by the parents and/or health-care professionals. The researcher read each child's referral letter in order to ensure that the referral included a question of social communication difficulties/ ASD. It is important to note that due to the complexity of the symptom presentations which are assessed within the clinic, many of the children included within this study had additional diagnoses, including ADHD, Rett Syndrome, or either general or specific learning difficulties.

## **2.4 The Assessment Procedure**

The assessments for the children included within this study consisted on average of 2.22 half day sessions. These were completed either in a single day, or over different half-day appointments. The average time in-between appointments for participants within this study was 28.1 days (sd=13.42).

The assessment follows a national recommended format, following NICE guidelines for autism diagnosis (Carr & O'Reilly, 2007). It is comprised of three main parts, including: a clinical interview to identify parents' questions and take a detailed developmental and family history; a child assessment conducted by different members of the MDT; and an MDT discussion to develop a diagnostic profile and formulate treatment recommendations. The child assessment varies depending on the presenting symptomatology. Parent and Teacher

reports of difficulties are assessed using questionnaire based assessments, for example the Strengths and Difficulties Questionnaire (SDQ: Goodman, 1997). A combination of standardised cognitive (e.g. the Weschler Intelligence Scale for Children – Fourth Edition: Weschler, 2003) and play-based assessments (e.g. the Autism Diagnostic Observations Schedules: Lord et al., 1989) are conducted with the child. When deemed necessary by the team, observations are conducted of the child within their school environment.

At the end of the final appointment, the family is given a short break, whilst the MDT confers regarding results and professional opinions. The assessment results are then fed back and discussed with the family; the family and child can decide whether or not the child remains in the room for this discussion. If the child prefers, he/she can stay outside of the room to play with the team's assistant Psychologist. The post-assessment discussion includes diagnostic opinions and an explanation of the child's developmental profile. Recommendations for management of care are also discussed with the family. A brief written summary is handed to the family at the end of the final appointment. The clinic aims to send out a full clinical report to a previously agreed circulation list including parents, school teachers and local health-care professionals, within a four-week period.

**Table 2.1***Demographic characteristics of the sample at each of the three time points*

	<b>Time 1 (n=37)</b>	<b>Time 2 (n=26)</b>	<b>Time 3 (n=11)</b>
<b>Age of Parent, years</b>			
Mean (SD)	43.36 (7.93)	43.04 (7.14)	44.73 (7.89)
Minimum/Maximum (range)	33 / 66 (33)	33 / 62 (29)	34 / 60 (26)
<b>Age of child at referral, years</b>			
Mean (SD)	9.73 (3.66)	9.42 (3.61)	10.57 (4.40)
Minimum/Maximum	3.25 / 16.67	4.42 / 16.67	5.42 / 16.67
Range	13.42	2.25	11.25
<b>Parent gender, n (%)<sup>*</sup></b>			
Male	4 (10.8)	3 (11.5)	1 (9.1)
Female	32 (86.5)	23 (88.5)	10 (90.9)
<b>Child gender, n (%)</b>			
Male	30 (81.1)	21 (80.8)	8 (72.7)
Female	7 (18.9)	5 (19.2)	3 (27.3)
<b>Parent Employment Status, n (%)<sup>*</sup></b>			
Full Time	5 (14.3)	3 (12.0)	-
Part Time	17 (48.6)	12 (48.0)	6 (54.5)
Homemaker/ Carer	12 (34.3)	10 (40.0)	5 (45.5)
Retired	1 (2.9)	-	-
<b>Marital Status, n (%)<sup>*</sup></b>			
Married	24 (66.7)	17 (65.4)	8 (72.7)
Single	6 (16.7)	5 (19.2)	3 (27.3)
Divorced	3 (8.3)	2 (7.7)	-
Cohabiting	2 (5.6)	1 (3.8)	-
Separated	1 (2.8)	1 (3.8)	-
<b>Educational Level, n (%)<sup>*</sup></b>			
GCSE or A-Level	11 (33.4)	9 (39.1)	5 (45.5)
Degree/Diploma	18 (54.5)	11 (47.8)	6 (54.5)
Postgraduate	4 (12.1)	3 (13.0)	-
<b>Ethnicity, n (%)<sup>*</sup></b>			
White British	23 (67.6)	18 (72.0)	9 (81.8)
White European	1 (2.9)	1 (4.0)	-
Asian British	3 (8.8)	2 (8.0)	-
British Other	6 (17.6)	3 (12.0)	1 (9.1)
Other	1 (2.9)	1 (4.0)	1 (9.1)

\* missing data from demographic questionnaire

## **2.5 Power Analysis**

A minimum required sample of 32 parents were calculated to be needed in order to reach adequate statistical power (Cohen's  $d=0.80$ ; Cohen, 1988); i.e. the ability of the analyses to be able to detect effects when they exist (Field, 2009). Prospective calculations were conducted using G-Power (version 3.1.2). This analysis was based on the first phase of this study (Moran et al., submitted); no other identified study has systematically measured parental understanding both pre and post diagnostic assessment using a validated tool. The first phase of this study achieved a response rate of 46% ( $n=59$ ) and a moderate effect size of  $r=0.50$  (Moran et al., submitted). This effect size relates to the correlation found between the PUN-Q and parental sense of competence (PSOC). Power for the current study was derived with regards to research question 1, which investigates the test-retest reliability of the PUN-Q between two time points using paired-samples t-tests. The 37 families recruited to the study at the time of writing this thesis led to a power of Cohen's  $d = .77$  at Time 1.

## **2.6 Design**

This was a prospective study with a longitudinal within-groups design. Parents of eligible children were followed up at three time points (see Figure 2.1). 'Time 1' was completed as soon as the patient's referral was accepted onto the waiting list and consent to join the study had been received; 'Time 2' marked the end of waiting list period and just before the first clinical appointment; 'Time 3' was completed immediately after the child and parent had attended their final assessment appointment at the clinic.

The families were also asked to complete questionnaires at a further time point – 'Time 4', which marked the receipt of the final clinical report, approximately six weeks following the family's final appointment. As a consequence of time constraints on recruitment, this final

time point was not included within the analyses for this thesis. Time 4 will be used within future analyses and in the write up of the full study for publication.

The intended gap between each time point was six to 10 weeks (see Figure 2.1 for the average number of weeks in-between time points). This varied between participants due to clinical considerations, including urgency of a child's referral, the number of appointments offered to the family, and parental time factors (e.g. the distance that they lived to the service and their availability for appointments).

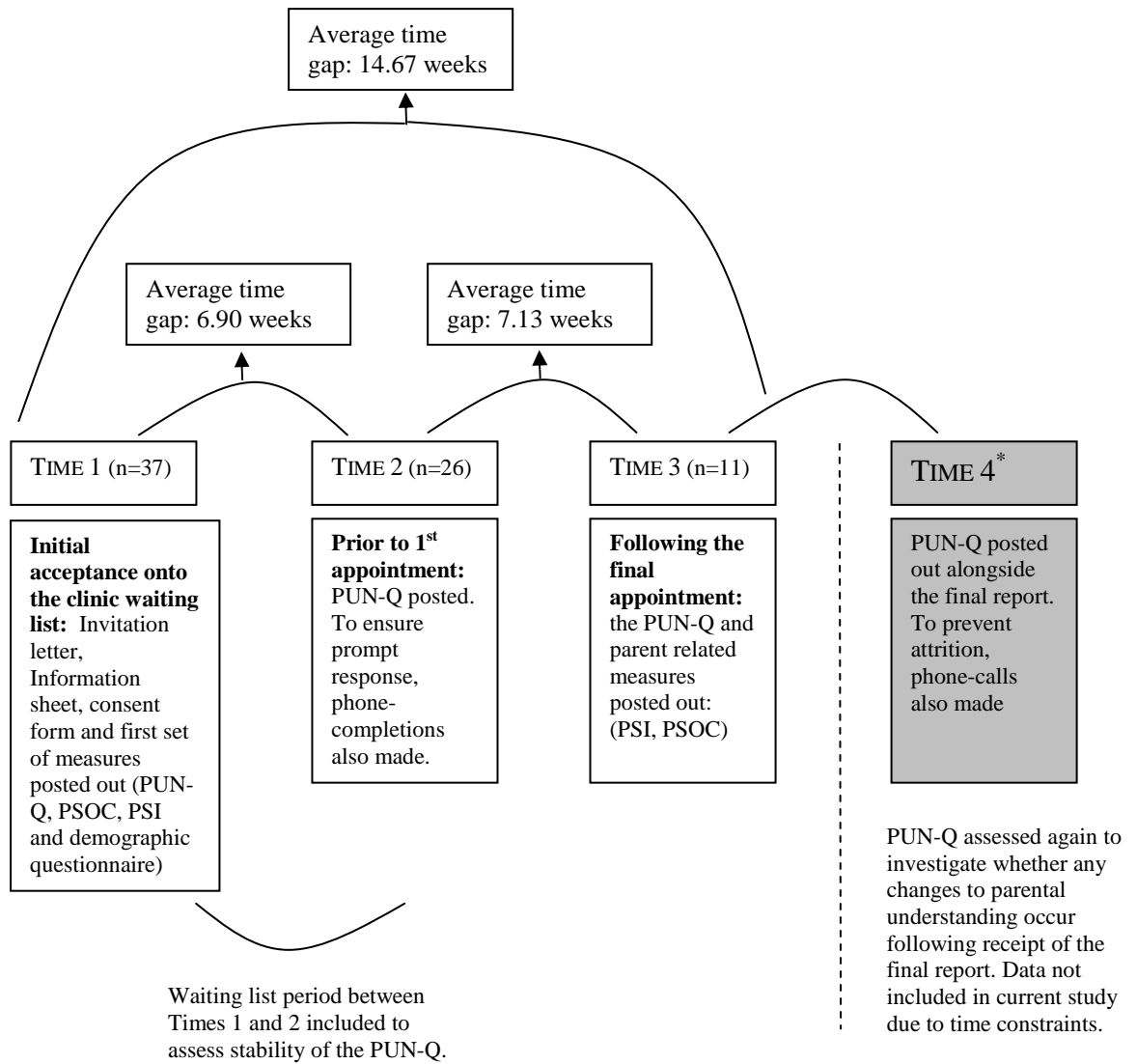
At the time of writing this thesis, 70.3% of parents (n=26) had completed 'Time 2' (the researcher was unable to contact one parent); and 29.7% (n=11) had completed 'Time 3' (one parent dropped out of the study at this stage as their child was referred to a different service).

## **2.7 Procedure**

Parents of children referred to the clinic for any form of social communication difficulties including queries regarding ASD (either diagnosed or undiagnosed) were invited by post to participate in the study. Parents were sent a pack consisting of a letter of invitation, a participants' information sheet, consent form and an initial battery consisting of four questionnaires (see Appendices 2 to 4 for copies of the documents included within the invitation pack): The Parental Understanding of Neurodisability Questionnaire (PUN-Q: Moran et al., submitted manuscript); The Parenting Stress Index – Short Form (PSI-SF: Abidin, 1995); The Parental Sense of Competence Questionnaire (PSOC: Johnston & Mash, 1989) and a demographic questionnaire (see Figure 2.1 for study design). As part of the clinic's routine clinical procedure parents were separately sent 'The Strengths and Difficulties Questionnaire' (SDQ: Goodman, 1997) and the 'Social Communication Questionnaire' (SCQ: Rutter et al., 2003); these two questionnaires were sent to parents before their first



appointment at the clinic and were returned to the clinicians involved with their child's assessment. The parental consent process allowed the researcher to gain access to these data from the two latter questionnaires for the purpose of the study.



**Figure 2.1:** Figure to show the prospective, longitudinal study design

Parents who were willing to participate in the study were asked to send their completed questionnaires and consent form back in a Freepost envelope. Approximately one to two weeks after the information packs were posted out, parents were telephoned by the researcher to verbally explain the study and to answer any questions that the parents may have had.

It was estimated that the questionnaire battery included in 'Time 1' (PUN-Q, PSOC, PSI, and demographic questionnaire) took parents approximately 20 minutes to complete. Approximately two weeks before the family's first appointment, families were contacted by post and asked to fill in the pre-assessment version of the PUN-Q for a second time (i.e. 'Time 2'). It was estimated that 'Time 2' took parents approximately five minutes to complete. This time point was included in order to allow an examination of the stability (i.e. test-retest reliability) of the PUN-Q over-time. In order to prevent attrition and due to the tight time-scales, families were also telephoned, and the pre-assessment PUN-Q completed, where necessary, over the phone with the researcher. Two weeks following their final appointment, families were sent four further questionnaires to complete and post back to the researcher: the post-assessment version of the PUN-Q; the PSI; the PSOC. It was estimated that filling in the questionnaire battery for 'Time 3' took parents approximately 20 minutes to complete.

The researcher telephoned the families at each time point to ensure that the questionnaire packs had been received and to answer any questions about the procedure that they may have had. This was to help ensure that the completed questionnaires were returned to the researcher as soon as possible, so that each of the time points remained independent of one another, without any temporal overlap.

## **2.8 Ethical Considerations**

Participation in all time points of the study was voluntary and involved informed and active (i.e. opt-in) written parental consent (see Appendix 4). Parental consent was obtained prior to Time 1 for all subsequent time points. Confidentiality was guaranteed; parents were informed that no-one except the researcher and Chief Investigator has access to their data. It was emphasised within all correspondence with the families that participation in this research would not affect the clinical care received; the clinicians involved with the families were not informed which families took part in the study and had no access to the data.

To ensure confidentiality of data, each family was identified by a unique participant ID number, which was allocated to them upon being sent the information pack and consent form; families were only identified by this number. Only the researcher and Chief Investigator had access to a master list, which linked ID numbers with the names of participating families. This master list was stored on a secure server and password protected. Completed questionnaires were kept in a locked office.

The first phase of the PUN-Q validation (Moran et al., submitted) was approved in 2011 by the NHS London Bloomsbury Research Ethics Committee (09/H0713/63). For the current study, a Notice of Substantial Amendment was granted in September 2014 by the same research ethics committee. Ethical approval was also gained for the current study from the Royal Holloway, University of London Ethics Committee in September 2014. Further approval was granted by the R&D office of the Institute of Child Health/ Great Ormond Street Hospital.

## 2.9 Measures

All measures were self-report questionnaires (see Figure 2.1 above for more information).

Please see Appendices 6 to 10 for copies of the measures.

### 2.9.1. The Parental Understanding of Neurodisability Questionnaire (PUN-Q: Moran et al., submitted).

This is a 13 item questionnaire using a 5-point Likert scale (ranging from strongly agree to strongly disagree). The first stage of this study developed this measure and conducted preliminary validation on a sample of 59 parents (Moran et al., submitted); the PUN-Q was developed for use with parents of children aged 0 to 18 years old. Results demonstrated high internal consistency (Cronbach  $\alpha=0.88$ ). A previously outlined factor analysis revealed three stable factors: '*post-assessment understanding*' (e.g. 'explanations that I have been given to explain my child's difficulties make a lot of sense to me'); '*insightful understanding*' (e.g. 'most of the time, I understand why my child behaves the way that s/he does'); and '*application of understanding*' (e.g. 'I know how to adjust what I do as a parent to take account of my child's difficulties'). Construct validity was shown through positive correlations with the PSOC ( $p<0.005$ ) and the PSI-SF ( $p<0.005$ ).

Within the current study the full 13 items of the PUN-Q (i.e. PUN-Q-13) were administered only at Time 3. Five items which make up 'Factor 1' of the PUN-Q were omitted from Times 1 and 2, due to their focus on '*post-assessment understanding*'. It was felt that these items, which pertained towards post-assessment understanding, were potentially confusing for parents to answer prior to their attendance within the clinic. Further, answers to these items if delivered prior to the assessment, may reflect parents' previous experiences with other services. This confusion could subsequently contaminate any measurement of changes to parental understanding which were influenced by the current diagnostic assessment.

Composite scores were created for the 8 ‘pre-assessment’ items at Times 1, 2 and 3 (PUN-Q-8), in addition to a total score for all 13 items at Time 3 (PUN-Q-13); higher PUN-Q-8 total scores indicated higher levels of parental understanding. Total scores were also created for each of the factors on the PUN-Q-8 at all time points and on the PUN-Q-13 at Time 3. Within the current study internal reliability was adequate for the PUN-Q at all time points: Time 1  $\alpha=.72$  (8 items); Time 2  $\alpha=.76$  (8 items); Time 3  $\alpha=.92$  (13 items).

### **2.9.2 The Parenting Sense of Competence Scale (Johnston & Mash, 1989).**

This is 17-item questionnaire using a six-point Likert scale (ranging from strongly agree to strongly disagree). This scale was used to measure parental self-perceptions of their parenting competence across two orthogonal constructs: ‘Efficacy’ and ‘Satisfaction’ (Johnston & Mash, 1989; Gilmore & Cuskelly, 2009). Scores were summed for each of the constructs separately, in addition to deriving a total composite score for parental sense of competence; higher scores indicated greater sense of competence.

Good levels of internal consistency have been previously reported for this measure (range  $r=0.75$  to  $r=0.88$ ) (e.g. Johnston & Mash, 1989; Lovejoy, Verda, & Hays, 1997; Ohan, Leung, & Johnston, 2000). Johnston and Mash (1989) demonstrated good concurrent validity, with scores negatively correlating with the internalising and externalising scales of the Child Behaviour Checklist (Achenbach, 1991).

This instrument was chosen for the current study as it is the most commonly used and standardised measure for measuring parenting self-efficacy. It was especially suitable for this study as it assesses parenting competence more generally, and is therefore appropriate for

parents of children with neurodevelopmental disorders. Within the current study internal reliability was good at both time points: Time 1  $\alpha=.88$ ; Time 3  $\alpha=.87$ .

### **2.9.3 The Parenting Stress Index – Short Form (PSI-SF: Abidin, 1995).**

This is a 36 item questionnaire using a five-point Likert scale (ranging from strongly agree to strongly disagree). Three subscales are measured: '*parental distress*' (distress resulting from difficulty coping, marital problems or restrictions due to caring for their child), '*difficult child*' (parental perceptions of children's self-regulatory abilities and their own ability to manage their child) and '*dysfunctional child-parent relationship*' (dissatisfaction from interactions with their child, viewing the child as a disappointment. Scores were summed for each of the subscales separately, in addition to deriving a total composite score for parental stress; higher scores indicated greater levels of stress.

Good construct validity has previously been demonstrated, with correlations in the expected direction against scales of depression and parental sense of competence. The PSI-SF has been shown to retain stability over a one year period, with correlations between the different subscales of between  $r=0.61$  to  $r=0.75$  over time (Haskett, Ahern, Ward, & Allaire, 2006). This scale was included within this study to measure parenting stress, as it has been used extensively within the literature, and previous studies have shown it to be a valid instrument for measuring parenting stress for parents of children experiencing symptoms of ASD and developmental delay (e.g. Hassall et al., 2005; Davis & Carter, 2008); Within the current study internal reliability was good at both time points: Time 1  $\alpha=.94$ ; Time 3  $\alpha=.91$ .

#### **2.9.4 Strengths and Difficulties Questionnaire (SDQ: Goodman, 1997).**

This is a 25-item questionnaire using a three-point Likert scale (ranging from not true to certainly true). The SDQ assesses five aspects of behaviours: emotional symptoms, conduct problems, hyperactivity/inattention, peer-problems and pro-social behaviour. A total difficulties composite score was derived; this did not include the pro-social behaviours subscale (Goodman, 2001). Total scores higher than 17 (range 0 to 40) were indicative of greater difficulties, falling within the 10<sup>th</sup> percentile in UK norms for SDQ scores (Meltzer et al., 2000).

The SDQ has been shown to be a reliable and well validated measure of children's emotional and behavioural symptoms. It is a widely used scale within clinical services, due to it being easy to administer with different versions for parents, children and teachers (Goodman, 2001).

The SDQ was used within this study as a measure child-related difficulties due to its' established validity and reliability (e.g. Goodman, 2001), in addition to the instrument being freely available and routinely administered within the study's clinic; use of this instrument therefore alleviated extra research burden for the participating parents. As a consequence of these data being collected by clinicians the raw data was not readily available, therefore it was not possible to calculate the internal consistency.

#### **2.9.5 Social Communication Questionnaire (SCQ: Rutter et al., 2003).**

This is a 40-item questionnaire, which is based on the Autism Diagnostic Interview – Revised (ADI-R: Lord, Rutter, & LeCouteur, 1994), and is used to ask parents about characteristic symptoms of ASD, either currently or across the lifetime. Items are rated dichotomously (0 or 1), where 1 indicates endorsement of a specific ASD symptom. Total scores were derived; scores of 15 or above are indicative of potential ASD or PDD.

The SCQ was used within the current study as it has been validated as a reliable screening questionnaire for ASD diagnoses (Berument et al., 1999), and is used routinely within the study's clinic; the use of this questionnaire therefore did not place any additional research burden upon the parents. The SCQ has been shown to have higher levels of sensitivity and specificity (0.86 and 0.78, respectively), when compared against two other widely used Autism screening parent-report questionnaires: the Social Responsiveness Scale (Constantino & Gruber, 2005) and the Children's Communication Checklist (Bishop, 1998), using a sample of 119 children aged between 9 and 13 years old (Charman et al., 2007). The sensitivity of the SCQ has been supported in further research which sampled a population cohort of children (Chandler et al., 2007). Further research has shown cross-cultural validity (e.g. Bolte, Holtmann, & Poustka, 2008). The SCQ data were collected by clinicians as part of the routine care offered by the clinic. Consequently, the raw data was not readily available, and it was not possible to calculate the internal consistency.

#### **2.9.6 Demographic Questionnaire (devised by Moran et al., submitted).**

This was a self-report demographic questionnaire, which parents completed at Time 1. It asked for information including the gender of the participating parent, parent age, family composition, gender of child, ethnicity, parent employment status, and highest level of parental education.

#### **2.10 Data Analyses**

All data were analysed using SPSS v.21; alpha levels were set at  $p < 0.05$ . All data were entered by the researcher. Data were screened prior to analyses following a procedure set out by Tabachnik and Fidell (2007) (e.g. ranges of each questionnaire checked for erroneous entries). Negatively phrased questions on both the PUN-Q and PSOC were reversed to ensure



that greater total scores on both measures reflected higher levels of parental understanding and sense of competence, respectively. Greater PSI scores reflected higher parenting stress levels. Total values were calculated for each measure, in addition to total values for the subscales included within the PSOC (*'Efficacy'* and *'Satisfaction'*) and PSI-SF (*'Parental Distress'*, *'Parent-Child Dysfunction'* and *'Difficult Child'*).

### **2.10.1 Missing Data.**

Two items of the PSI-SF had some missing data. This may reflect the PSI-SF being included last within the questionnaire battery. As a consequence of the relatively few missing cases (less than the 5% cut-off stipulated to be important: Tabachnik and Fidell, 2007), and due to these cases being missing from a well-validated scale, it was deemed sufficient to replace these items with the whole-group mean for that item, at that particular time point (Tabachnik and Fidell, 2007). Total scores were recalculated to take replaced values into account. No items had more than 5% of values missing, therefore no further investigations regarding missing data were conducted (Tabachnik and Fidell, 2007).

### **2.10.2 Outlier Analysis.**

Outliers represent data values that deviate from the other observations. They are important to detect as they may indicate difficulties within the data and lead to inaccurate error rates of statistical estimates, causing potentially erroneous results (Field, 2009). Univariate outliers can be checked by assessing the variability of standardised z-scores; z-scores greater than 3.29 indicate the presence of an outlier (Tabachnick and Fidell, 2007). Z-scores were calculated for total and subscale scores at each time point for all of the measures. Results indicated no univariate outliers within the data (see Table 2.2 for minimum and maximum values).

**Table 2.2***Minimum and Maximum Z-Score Totals*

	Time 1		Time 2		Time 3	
	(n=37)		(n=26)		(n=11)	
	Min.	Max.	Min.	Max.	Min.	Max.
<b>PUN-Q</b>						
Total Score PUN-Q-8	-1.33	2.99	-1.79	2.17	-1.68	1.31
Total Score PUN-Q-13	-	-	-	-	-1.70	1.20
Insightful Understanding	-1.58	2.92	-1.42	2.21	-1.35	1.35
Post assessment understanding	-	-	-	-	-1.74	1.08
Application of understanding	-1.94	2.60	-2.12	1.63	-1.84	1.21
<b>PSOC</b>						
Total Score	-2.09	1.83	-	-	-2.12	1.17
Efficacy	-1.96	1.94	-	-	-1.65	1.42
Satisfaction	-1.81	2.05	-	-	-2.55	.98
<b>PSI-SF</b>						
Total Score	-2.59	1.81	-	-	-1.31	1.98
Parental Distress	-1.78	1.74	-	-	-1.98	1.51
Parent-Child Dysfunction	-1.97	1.94	-	-	-.93	2.28
Difficult Child	-2.74	1.42	-	-	-1.73	1.48

**2.10.3 Normality of data.**

The small sample sizes for each time point resulted in non-parametric data. In order to increase the statistical power and still be able to answer the study's research questions, the following analyses utilised bias corrected and accelerated bootstrapping confidence intervals (Wichmann & Hill, 2001).

Bootstrapping is a non-parametric procedure which does not therefore assume normality of data (Preacher & Hayes, 2004). It is utilised when parametric assumptions for data are in

doubt. This can occur due to small sample sizes; a large sample size is considered  $>30$  for bootstrapping purposes, whilst a sample of  $n \geq 8$  is considered adequate for reliable results (Zhu, 1997). This method involves repeated ‘resampling’, with replacement, from the study’s dataset (at least 1000 times is advised), thus creating phantom samples. The more bootstraps that are conducted, the greater probability there is that the bootstrapped confidence intervals (CI) represent valid results (Davidson & McKinnon, 2001). These bootstrapped sampling distributions are then used as non-parametric approximations of the study’s sampling distribution (they create an approximation for a normal distribution). This process enables the construction of robust estimates of standard errors and CI for smaller sample sizes with non-parametric distributions (see Preacher & Hayes, 2004).

The bias corrected and accelerated method derives CI with a higher level of accuracy (Wichmann & Hill, 2001). These CI which are based on an approximation of the sampling distribution, do not need to be symmetrical, and therefore are not prone to the inaccuracies and power difficulties prevalent with the use of ordinary CI (Efron and Tibshirani, 1993). Bootstrapping has been shown to increase the statistical power of the analyses, without inflating the Type-I or Type-II error rates (see MacKinnon et al., 2002; Peacher and Hayes, 2004).

For the purposes of this study, 5000 bias corrected and accelerated bootstrapped CI were derived for all analyses, with significance levels set at 95%. This number of bootstrapped re-samples was considered sufficient, as results did not substantially vary when repeated (Davidson & McKinnon, 2001). Significant effects are present when CI do not include 0; in cases of conflict with the non-bootstrapped p-values, the bootstrapped CI were favoured (p-values are reported alongside the CI).

#### **2.10.4 Multicollinearity.**

Multicollinearity is considered a problem when high correlations exist ( $r > 0.90$ ) between variables (Field, 2009). Multicollinearity suggests that items are redundant as a consequence of measuring the same latent variable; such variables would not therefore be necessary for the analyses. A correlation matrix was conducted to assess whether any inter-item correlations were above  $r = .90$  for the PUN-Q-8 at Time 1 and Time 2, and for the PUN-Q-13 at Time 3 (see Appendix 12). No correlations were shown to violate multicollinearity assumptions at Times 1 or 2. Two correlations were greater than .90 at Time 3. This is discussed within the Discussion chapter as a limitation for the PUN-Q-13 data at this time point; results should therefore be interpreted with caution for the Time 3 data.

#### **2.10.5 Potential Confounding Variables.**

Categorical demographic variables were split into two groups based on median values. Independent samples t-tests were conducted to examine whether the Time 1 PUN-Q-8 total score significantly differed between the two groups for the following variables: parental age, age of child at first appointment, employment status, and educational level, and the number of days parents waited between receipt of referral and their child's first appointment. Where results showed that the total PUN-Q-8 score did not differ based on a specific variable, that variable was not included as a covariate within the remaining analyses.

#### **2.10.6 Research Question 1: Construct Validity.**

Construct validity was examined at 'Time 1' by conducting Pearson's Correlations between the PUN-Q-8 total score and subscale scores, and both the total and subscales scores for the PSI-SF and PSOC. Additional Partial Correlations examined post-assessment associations between both the PUN-Q-8 and the PUN-Q-13 total scores at 'Time 3' with the concurrent

‘Time 3’ values for the PSOC and PSI-SF; these analyses controlled for baseline scores of the PUN-Q-8, PSOC and PSI-SF. Correlations between the subscales were not examined at Time 3, due to the smaller sample size decreasing the statistical power to detect effects.

#### **2.10.7 Research Question 2: Testing the stability of the PUN-Q-8.**

Paired samples t-tests were conducted using ‘Time 1’ and ‘Time 2’ total scores for the PUN-Q-8 in order to assess the stability of the PUN-Q-8 over time. This analysis formed part of the prospective validation of the PUN-Q-8 following the guidelines set out by Guyaatt and colleagues (1987), in order to assess the reliability over time of the PUN-Q-8 without potential intervention effect of the diagnostic assessment. The average time period between these time points was 6.9 weeks.

#### **2.10.8 Research Question 3: Sensitivity of the PUN-Q to measuring change.**

Post assessment analyses relied on the measurements collected at ‘Time 3’. Due to the small sample size for this time point in particular, even with the increased power afforded by the bootstrapping method, the following analyses are preliminary and should be interpreted with caution.

Total scores for the PUN-Q-8 were compared between ‘Time 1’ and ‘Time 3’ using Paired Samples T-Tests. Analyses were conducted for the total scores, and separately for both of the PUN-Q-8 factors (insightful understanding and application of understanding).

Cohen’s ‘d’ was calculated to estimate the effect size, or magnitude of change, in mean scores between the PUN-Q-8 total scores at Time 1 and Time 3. When interpreting these scores, 0.2 is regarded as ‘small’, 0.5 is regarded as ‘moderate’, 0.8 is regarded as ‘large’ (Cohen, 1992).

A step-wise Multiple Regression analysis assessed the association between the Time 1 total PUN-Q-8 score and the Time 3 PUN-Q-8 total score; the 'Time 3' PUN-Q-8 total score was included within the analysis as the dependent variable. The first step of the regression included the Time 1 PUN-Q-8 total score, the second step included the five items that make up the 'post assessment' factor of the PUN-Q-13. The final step included Time 1 total scores for the PSOC and PSI-SF, in addition to the SDQ total score; the SCQ total score was not included due to the binary nominal nature of these data.

#### **2.10.9 Effects of the Diagnostic Assessment.**

The ability of the intervention to affect change for levels of 'parenting stress' (measured by the PSI-SF) and 'parental sense of competence' (measured by the PSOC) was examined using paired samples t-tests, between 'Time 1' and 'Time 3' total scores for each measure respectively. Analyses were then repeated for the different subscales for each of the scales. These secondary analyses were conducted in order to be able to compare any effect of the intervention shown for the PUN-Q-8 total score.

#### **2.10.10 Research Question 4: Exploration of the relationship between the PUN-Q and child difficulties**

This thesis examined whether the Time 1 PUN-Q 8 total score was associated with the complexity of a child's difficulties.

##### *2.10.10.1 Child Total Difficulties.*

A Pearson's Correlation was conducted to examine the relationship between the PUN-Q-8 total score and the SDQ total difficulties score. Comparison correlations were conducted

between the SDQ total difficulties score and the Time 1 total scores for both the PSOC and PSI-SF measured at baseline.

#### *2.10.10.2 Child Social Communication Difficulties*

An Independent Samples T-Test was conducted to examine whether there were differences in the Time 1 total PUN-Q-8 score depending on the level of child social communication difficulties (as measured by the SCQ). The binary nominal nature of these data did not allow correlations to be conducted. Comparison analyses were conducted with the Time 1 PSOC and PSI-SF total scores.

## **CHAPTER 3: RESULTS**

### **3.1 Potential Confounding Variables**

Table 2.1 within the Methodology Chapter showed a break-down of the demographic information for the data that was available within the sample. A median split was applied and Independent Sample T-Tests conducted to examine the potential effect of the following variables upon the data for Time 1 total scores for the PUN-Q-8 (i.e. parental understanding), PSOC (i.e. parental sense of competence) and PSI-SF (i.e. parenting stress).

#### **3.1.1 Parental Age.**

Seventeen parents (47.2%) were aged between 33-41 years old (median age=42.0 years). Results showed no significant differences between the age groups for total scores on the PUN-Q-8 ( $t(34)=1.16$ ,  $p=.26$ ; BCa CI: -1.19 to 4.61), PSOC ( $t(34)=1.33$ ,  $p=.19$ ; BCa CI: -3.70 to 15.32) or PSI-SF ( $t(34)=-1.35$ ,  $p=.19$ ; BCa CI: -27.36 to 4.95).

#### **3.1.2 Child's Age at Referral.**

Fifty percent of children were aged between 8.92 and 16.67 years at point of referral (median age=9.50 years). Results showed no significant differences between the child-age groups for total scores on the PUN-Q-8 ( $t(30)=1.03$ ,  $p=.31$ ; BCa CI: -1.31 to 4.12), PSOC ( $t(30)=.94$ ,  $p=.35$ ; BCa CI: -5.45 to 14.46) or PSI-SF ( $t(30)=-1.16$ ,  $p=.26$ ; BCa CI: -25.88 to 5.93).

#### **3.1.3 Parental Employment Status.**

Parents were divided into those who were employed (either part or full time) and those who were fulltime caregivers or home-makers; 62.9% ( $n=22$ ) of the sample were in either part or full time employment. Results showed no significant differences between the two employment groups for total scores on the PUN-Q-8 ( $t(32)=.17$ ,  $p=.87$ ; BCa CI: -3.80 to



3.73), PSOC ( $t(32)=.10$ ,  $p=.92$ ; BCa CI: -7.80 to 8.87) or PSI-SF ( $t(32)=-.46$ ,  $p=.65$ ; BCa CI: -21.94 to 14.96).

#### **3.1.4 Parental Education Level.**

Parents were divided into those who were educated up to A-Level education level and those who were educated at degree level or higher (11 parents (33.3%) were educated up to A-Levels). Results showed no significant differences between the two education groups for total scores on the PUN-Q-8 ( $t(31)=.31$ ,  $p=.76$ ; BCa CI: -3.67 to 3.86), PSOC ( $t(31)=.28$ ,  $p=.78$ ; BCa CI: -9.04 to 12.41) or PSI ( $t(31)=.07$ ,  $p=.94$ ; BCa CI: -17.07 to 17.96).

#### **3.1.5 Previous Child Neurodevelopmental Diagnosis.**

Parents were split between those whose child had previously received a neurodevelopmental diagnosis ( $n=21$ ), and those who had not ( $n=12$ ). Results showed no significant between-group differences for total scores on the PUN-Q-8 ( $t(31)=.33$ ,  $p=.74$ ; BCa CI: -3.64 to 4.19), PSOC ( $t(31)=-.97$ ,  $p=.34$ ; BCa CI: -14.13 to 3.83) or PSI-SF ( $t(31)=.92$ ,  $p=.37$ ; BCa CI: -7.00 to 23.16).

#### **3.1.6 Days Waiting between Referral and First Appointment.**

Parents waited between 52 and 151 days between acceptance of referral and their child's first appointment (average=102.69 days; median=109.50 days). Results showed no significant differences for total scores on the PUN-Q-8 ( $t(24)=1.36$ ,  $p=.19$ ; BCa CI: -1.14 to 5.31), PSOC ( $t(24)=.19$ ,  $p=.85$ ; BCa CI: -10.96 to 12.05) or PSI ( $t(24)=.17$ ,  $p=.87$ ; BCa CI: -16.00 to 20.20), based on number of days waiting for the first appointment.

As a consequence of the non-significant effects shown for these variables on total Time 1 PUN-Q-8 scores (i.e. parental understanding), the following analyses were conducted without including these variables as covariates.

### **3.2 Research Question 1: Time 1 Construct Validity for the PUN-Q**

#### **3.2.1 PUN-Q-8 and parenting self-efficacy (PSOC).**

Table 3.1 above shows the Pearson's correlations with bias corrected and accelerated Confidence Intervals (BCa CI) between Time 1 total scores and subscale scores for the PUN-Q-8, PSOC and PSI-SF. Significant positive correlations were shown between the PUN-Q-8 and PSOC total scores ( $r=.42$ ,  $p=.01$ ; BCa CI: .17 to .64), in addition to the total PUN-Q-8 score and the 'parenting efficacy' subscale of the PSOC ( $r=.44$ ,  $p=.01$ ; BCa CI: .20 to .65). A significant association was indicated in the BCa CI between the PUN-Q total score and the 'parenting satisfaction' subscale of the PSOC ( $r=.29$ ,  $p=.08$ ; BCa CI: .02 to .53). These results suggest that a higher PUN-Q-8 score (i.e. parental understanding) is associated with a higher score on PSOC (i.e. parenting self-efficacy), as theoretically predicted.

The 'application of understanding' PUN-Q-8 subscale was significantly positively correlated with the PSOC total score ( $r=.47$ ,  $p<.01$ ; BCa CI: .20 to .70), in addition to the PSOC 'parenting efficacy' ( $r=.44$ ,  $p=.01$ ; BCa CI: .18 to .66) and 'parenting satisfaction' subscales ( $r=.41$ ,  $p=.01$ ; BCa CI: .20 to .70). These results suggest that as predicted, higher parental perceptions of their ability to practically apply understanding to their care-giving is associated with higher parenting self-efficacy scores.

No significant associations were shown between the 'insightful understanding' subscale of the PUN-Q-8 and the total PSOC score ( $p=.10$ ), or either the PSOC 'parenting satisfaction' ( $p=.43$ ), or 'parenting efficacy' subscales ( $r=.33$ ,  $p=.04$ ; BCa CI: -.04 to .66). These results suggest that the PUN-Q-8 'insightful understanding' factor is not associated with parental sense of competence, within this sample.

**Table 3.1***Pearson's Correlations matrix between PUN-Q-8, PSOC and PSI-SF at Time 1*

	1	2	3	4	5	6	7	8	9
1. PUN-Q total score									
BCa CI: Lower/Upper	-								
Std. Error	-								
2. PUN-Q: Insightful understanding									
BCa CI: Lower/Upper	.80/.96***	-							
Std. Error	.04	-							
3. PUN-Q: application of understanding									
BCa CI: Lower/Upper	.67/.91***	.14/.76**	-						
Std. Error	.06	.16	-						
4. PSOC total score									
BCa CI: Lower/Upper	.17/.64	-.09/.60	.20/.70**	-					
Std. Error	.12	.18	.12	-					
5. PSOC: Efficacy									
BCa CI: Lower/Upper	.20/.65**	-.04/.66*	.18/.66**	.88/.97***	-				
Std. Error	.12	.17	.13	.02	-				

	1	2	3	4	5	6	7	8	9
6. PSOC: Satisfaction									
BCa CI: Lower/Upper	.02/.53	-.17/.41	.12/.68*	.77/.94***	.39/.80***	-			
Std. Error	.13	.16	.16	.04	.11	-			
7. PSI total score									
BCa CI: Lower/Upper	-.62/-.18**	-.60/-.03*	-.61/-.16*	-.88/-.64***	-.87/-.60***	-.81/-.39**	-		
Std. Error	.11	.15	.12	.06	.07	.11	-		
8. PSI: Parental distress									
BCa CI: Lower/Upper	-.56/-.15*	-.53/.04	-.60/-.15*	-.85/-.50***	-.84/-.48**	-.79/-.32***	.73/.92**	-	
Std. Error	.11	.15	.12	.09	.09	.12	.05	-	
9. PSI: Parent-child dysfunction									
BCa CI: Lower/Upper	-.49/-.16	-.50/-.06	-.49/-.04	-.76/-.37***	-.77/-.34**	-.69/-.17**	.69/.92*	.27/.77***	-
Std. Error	.10	.13	.12	.11	.11	.13	.05	.13	-
10. PSI: Difficult child									
BCa CI: Lower/Upper	-.67/.09	-.64/.16	-.62/.03	-.80/-.41***	-.79/-.36***	-.75/-.25*	.65/.90**	.19/.73**	.27/.72**
Std. Error	.18	.19	.17	.11	.11	.13	.07	.14	.11

\*\*\*p<.001; \*\*p<.01; \*p<.05 (p-values based on non-bootstrapped estimates)

NB// PUN-Q-8: parental understanding; PSOC: parenting sense of competence; PSI: parenting stress

### 3.2.2 PUN-Q-8 and parenting stress (PSI-SF).

Significant negative Pearson's correlations were shown between the total scores for the PUN-Q-8 and PSI-SF ( $r=-.43$ ,  $p=.01$ ; BCa CI:  $-.62$  to  $-.18$ ). There was a significant negative correlation between the PUN-Q-8 total score and both the PSI-SF 'parenting distress' ( $r=-.37$ ,  $p=.02$ ; BCa CI:  $-.56$  to  $-.15$ ) and 'parent-child dysfunctional relationship' subscales ( $r=-.33$ ,  $p=.05$ ; BCa CI:  $-.49$  to  $-.16$ ). No significant association was shown between the PUN-Q-8 total score and the 'difficult child' subscale of the PSI-SF ( $r=-.36$ ,  $p=.03$ ; BCa CI:  $-.67$  to  $.09$ ). These results suggest that as hypothesised, higher levels of the PUN-Q-8 (i.e. parental understanding) are associated with lower parenting stress levels.

The PUN-Q-8 'insightful understanding' subscale was significantly, negatively associated with the total PSI-SF score ( $r=-.35$ ,  $p=.03$ ; BCa CI:  $-.60$  to  $-.03$ ), in addition to the PSI-SF 'parent-child dysfunction' subscale ( $r=-.29$ ,  $p=.08$ ; BCa CI:  $-.50$  to  $-.06$ ). No significant associations were shown between 'insightful understanding' and the other PSI-SF subscale scores. There was a significant negative association between the PUN-Q-8 'application of understanding' subscale and the total PSI-SF score ( $r=-.40$ ,  $p=.02$ ; BCa CI:  $-.61$  to  $-.16$ ).

Significant associations were also shown between the 'application of understanding' subscale and both the 'parenting distress' ( $r=-.39$ ,  $p=.02$ ; BCa CI:  $-.60$  to  $-.15$ ) and 'parent-child relationship dysfunction' PSI-SF subscales ( $r=-.27$ ,  $p=.11$ ; BCa CI:  $-.49$  to  $-.04$ ). No significant association was shown between the PUN-Q-8 'application of understanding' subscale and the PSI-SF 'difficult child' subscale ( $r=-.32$ ,  $p=.06$ ; BCa CI:  $-.62$  to  $.03$ ). These results suggest that higher levels of parents' perceived ability to apply understanding to their child are associated with lower levels of both parenting distress and difficulties within the parent-child relationship.

The significant associations shown between the total score for the PUN-Q-8 and both the PSOC and PSI were in the expected directions as theoretically predicted. These results therefore support the hypotheses and provide further validity to the shortened PUN-Q-8 measure, which supports the construct validation of the PUN-Q-13 that was demonstrated within the first phase of this study (Moran et al., submitted). In particular, they show that at Time 1, before the new assessment episode has started, parental understanding of their child's neurodisability symptoms correlate positively with parental sense of competence and negatively with parenting stress.

### **3.2.3 Time 3 Construct Validity for the PUN-Q.**

Table 3.2 shows the Partial correlations with bias corrected and accelerated Confidence Intervals (BCa CI) between the total scores for the PUN-Q-8, PUN-Q-13, parenting sense of competence (PSOC) and parenting stress (PSI-SF) at Time 3. Results showed no concurrent significant associations between either the PUN-Q-8 or PUN-Q-13 with total scores for either the PSI-SF score ( $r = -.11$ ,  $p = .79$ ; BCa CI:  $-.90$  to  $.62$ ;  $r = .08$ ,  $p = .86$ ; BCa CI:  $-.91$  to  $.90$ , respectively) or PSOC ( $r = -.16$ ,  $p = .70$ ; BCa CI:  $-1.00$  to  $1.00$ ;  $r = -.39$ ,  $p = .34$ ; BCa CI:  $-1.00$  to  $.99$ , respectively). These results are contrary to the a-prior hypotheses, which stated that post assessment, the PUN-Q would be positively associated with the PSOC and negatively associated with the PSI-SF. Interestingly, no significant correlation was shown at Time 3 between the PSOC or PSI-SF total scores. These results need to be interpreted with caution due to the smaller sample size at Time 3 and will be discussed further within the Discussion chapter.

**Table 3.2**

*Associations between the PUN-Q, parenting sense of competence and parenting stress at Time 3*

	1	2	3	4
1. PUN-Q-8 total score				
BCa CI: Lower/Upper	-			
Std. Error	-			
2. PUN-Q-13 total score				
BCa CI: Lower/Upper	.18/1.00**	-		
Std. Error	.20	-		
3. PSOC total score				
BCa CI: Lower/Upper	-1.00/1.00	-1.00/.99	-	
Std. Error	.54	.50	-	
4. PSI-SF total score				
BCa CI: Lower/Upper	-.90/.62	-.91/.90	-1.00/.98	-
Std. Error	.47	.53	.53	-

NB// Partial Correlations controlled for Time 1 total scores on the PUN-Q-8, PSOC and PSI-SF

\*\*  $p < .01$

### 3.3 Research Question 2: PUN-Q test-retest reliability between Times 1 and 2

Table 3.3 shows the means and standard deviations for the PUN-Q at the three different time points. A paired samples t-test with BCa CI showed no significant differences between the PUN-Q-8 total scores at Time 1 and Time 2 ( $t(25) = -.39$ ,  $p = .70$ ; BCa CI: -1.89 to 1.34). No significant differences were shown between Time 1 and Time 2 for either the 'insightful understanding' factor ( $t(25) = -.33$ ,  $p = .74$ ; BCa CI: -1.35 to 1.00), or for the 'application of understanding' factor ( $t(25) = -.28$ ,  $p = .78$ ; BCa CI: -.97 to .65). These non-significant

differences between the two pre-assessment PUN-Q-8 total scores at Time 1 and Time 2 suggest test-retest reliability during the ‘non-interventionist’ waiting period.

### **3.3.1 Behaviour of the PUN-Q pre and post assessment (i.e. Times 1 and 3).**

Pearson’s Correlations further explored the behaviour of the PUN-Q over time, by examining the association between the PUN-Q-8 total scores at Time 1 and Time 3. Results showed significant positive correlation between the two time points ( $r=.77$ ,  $p=.01$ ; BCa CI: .52 to .93). Analyses were repeated between Time 1 and Time 3 for the PSOC and PSI-SF total scores. Results showed significant positive associations between the two time points for both the PSOC ( $r=.94$ ,  $p<.001$ ; BCa CI: .84 to .99) and PSI-SF ( $r=.85$ ,  $p=.001$ ; BCa CI: .55 to .96). Consequently, these results suggest that whilst the concurrent relationship between the measures has disappeared at Time 3, these measures each show consistent associations between the pre-and-post assessment time points.

### **3.4 Research Question 3: Sensitivity of the PUN-Q to measure change**

Paired Samples T-Tests with BCa CI were conducted to compare the PUN-Q-8 total scores at ‘Time 1’ and ‘Time 3’ (see Table 3.3 for the means and standard deviations). Cohen’s ‘d’ was also calculated to assess the magnitude of change in total PUN-Q-8 scores between Times 1 and 3. Results showed a significant difference ( $t(10)=-3.46$ ,  $p=.01$ ; BCa CI: -7.00 to -2.00); PUN-Q-8 scores were significantly higher levels at Time 3 (i.e. post diagnostic assessment) in comparison to Time 1 pre-assessment scores (Cohen’s ‘d’=0.51, indicating a moderate effect size). Significantly higher scores were shown at Time 3 in comparison to Time 1 for both the PUN-Q-8 ‘insightful understanding’ ( $t(10)= -3.57$ ,  $p=.01$ ; BCa CI: -4.09 to -1.36; Cohen’s ‘d’=.48, indicating a small to moderate effect size), and ‘application of understanding’ subscales ( $t(10)=-2.07$ ,  $p=.07$ ; BCa CI: -3.00 to -.18; Cohen’s ‘d’=.46, indicating a small to moderate effect size).



**Table 3.3***Means and Standard Deviations for the measures at the different time points.*

		<b>Time 1 (n=37)</b>	<b>Time 2 (n=26)</b>	<b>Time 3 (n=11)</b>
<b>PUN-Q</b>	Total score (PUN-Q-8)	19.84 (4.40)	20.58 (4.79)	22.64 (6.34)
	Post-assessment PUN-Q 13	-	-	39.82 (9.29)
	Insightful understanding	9.56 (2.89)	9.92 (2.76)	11.00 (2.97)
	Application of understanding	10.27 (2.21)	10.66 (2.67)	11.64 (3.61)
<b>PSOC</b>	Total score	66.78 (13.76)	-	67.82 (12.09)
	Efficacy	35.08 (8.70)	-	34.36 (7.47)
	Parenting Satisfaction	31.71 (6.47)	-	33.45 (5.66)
<b>PSI-SF</b>	Total score	111.94 (24.85)	-	109.71 (20.81)
	Parenting distress	31.22 (10.82)	-	33.18 (9.16)
	Parent-child dysfunction	35.17 (9.71)	-	33.89 (8.83)
	Difficult child	45.56 (9.50)	-	42.64 (9.03)

NB// Standard deviations are in parentheses.

In order to explore whether the assessment may account for the increase PUN-Q-8 total scores, a step-wise multiple linear regression was conducted, with the Time 3 PUN-Q-8 total score as the dependent variable (see Table 3.4 for more information, including the BCa CI). Results showed a significant association between the Time 1 and Time 3 PUN-Q-8 total scores within step 1 of the model (BCa CI: .68 to 1.65). The strength of this association

decreased (as shown by the BCa CI being nearer to zero), but retained significance, upon the inclusion of the five items comprising the ‘post-assessment understanding’ factor of the PUN-Q-13 within step 2 of the model (BCa CI: .33 to 1.36). The association retained significance upon the introduction of the PSOC, PSI-SF and SDQ measures into the model at ‘step 3’ (BCa CI: .86 to 1.87), suggesting that the other perceived parental factors and child difficulties do not account for the post-assessment increase to parental understanding.

The reduction in the strength of the association between the PUN-Q-8 total scores between Time 1 and Time 3, upon the introduction of the PUN-Q-13 ‘post assessment understanding’ items into the model, suggests that the effects of the assessment accounted for part of the variance within this relationship; i.e. part of the increase shown for parental understanding.

**Table 3.4***Step-wise multiple linear regression model*

	Adjusted R2	BCa Standard Error	BCa Confidence Intervals	
			Lower	Upper
			Limit	Limit
<b>Model 1</b>	.55*			
Time 1: PUN-Q-8		.35	.68	1.65
<b>Model 2</b>	.67**			
Time 1: PUN-Q-8		.41	.33	1.36
Post-assessment understanding***		.50	-.00	1.63
<b>Model 3</b>	.83*			
Time 1: PUN-Q-8		2.10	.86	1.87
Post-assessment understanding***		3.66	-27.27	15.51
SDQ		2.28	-1.02	9.69
PSOC		.67	-2.90	.70
PSI-SF		.56	-2.09	.09

\* p&lt;.05; \*\*p&lt;.001

\*\*\* PUN-Q-13 ‘post assessment understanding’ subscale measured at Time 3

NB// BCa CI and standard errors are reported instead of Beta values

### **3.4.1 Effects of the Diagnostic Assessment: Parental Self-efficacy and parenting stress.**

Table 3.3 shows the means and standard deviations for the PSOC and PSI-SF at Time 1 and Time 3. Paired samples t-tests with BCa CI were conducted to compare mean pre-and-post assessment scores for the total PSOC and PSI-SF scores, respectively. Results showed no significant differences between the Time 1 and Time 3 scores for either the PSOC ( $t(10)=-1.64$ ,  $p=.14$ ; BCa CI: -4.09 to .27) or PSI-SF ( $t(10)=.70$ ,  $p=.49$ ; BCa CI: -5.12 to 9.62). These preliminary results within this small sample show no evidence to suggest that the clinical assessment process helped to enhance parenting sense of competence or to decrease parenting stress levels.

## **3.5 Research Question 4: Association between the PUN-Q-8 and Child Difficulties**

### **3.5.1 The PUN-Q-8 and Child Emotional and Behavioural Difficulties.**

Pearson's Correlations were conducted to explore the association between the Time 1 PUN-Q-8 total score and the child's total difficulties (as measured by the SDQ). Comparison correlations were conducted for Time 1 PSOC and PSI-SF total scores. Child total difficulties within this sample ranged from 15 to 37 ( $n=16$ ). Twelve children scored above 17, which is the threshold for 'abnormally high' difficulties (Goodman, 1997) (see Table 3.5). Results showed no significant association between the PUN-Q-8 total score and total child difficulties ( $r=-.31$ ,  $p=.24$ ; BCa CI: -.81 to .30). Significant associations were shown between total child difficulties and total scores for both the PSOC ( $r=-.65$ ,  $p=.006$ ; BCa CI: -.84 to -.40), and PSI-SF ( $r=.63$ ,  $p=.009$ ; BCa CI: .14 to .28). This indicates that within this sample, higher levels of child difficulties were associated with lower perceived parental self-efficacy and higher levels of parenting stress. These associations contrasted to the non-significant association shown

between child difficulties and the total PUN-Q-8 score (i.e. parental understanding of their child's neurodisability symptoms).

**Table 3.5**

*Published thresholds of the SDQ and levels in the current sample*

	Published SDQ Categories*			Current Sample	
	Normal	Borderline	Abnormal	Mean (sd)	Range
Total Difficulties	0-13	14-16	17-40	23.19 (6.50)	15-37
Emotional Symptoms	0-3	4	5-10	6.38 (3.28)	0-10
Conduct Problems	0-2	3	4-10	3.75 (2.62)	0-9
Hyperactivity Difficulties	0-5	6	7-10	7.69 (2.41)	1-10
Peer Problems	0-2	3	4-10	5.38 (1.67)	2-8
Pro-social Behaviour	6-10	5	0-4	5.00 (2.00)	1-9

\* Goodman (1997)

### 3.5.2 The PUN-Q-8 and Child Social Communication Difficulties.

Independent Samples T-Tests examined the association between the child's social communication difficulties and the Time 1 PUN-Q-8 total score. Comparison analyses were conducted for the PSOC and PSI-SF measures. The threshold for suspecting ASD using the SCQ measure is a score of 15 (Berument et al., 1999), however only four out of the 15 participants for whom this information was available were sub-threshold. Berument and colleagues (1999) suggest that other thresholds can be acceptable depending on the population being investigated. Accordingly, a median split was applied to the data. Results showed no significant differences between levels of child social communication difficulties for total scores on the PUN-Q-8 ( $t(13)=-1.33$ ,  $p=.21$ ; BCa CI: -5.99 to 1.30), PSOC ( $t(13)=-1.16$ ,  $p=.27$ ; BCa CI: -24.99 to 6.01) or PSI-SF ( $t(13)=-.98$ ,  $p=.35$ ; BCa CI: -9.92 to 36.79).

## **CHAPTER 4: DISCUSSION**

### **4.1 Summary of findings**

Using data collected over three time points, this thesis aimed to prospectively examine the reliability and validity for a newly developed measure (the PUN-Q) (Moran et al., submitted), which aims to examine parental understanding of their child's neurodisability symptoms. Novel to this thesis was the examination of the PUN-Q both prior to and following a neurodevelopmental diagnostic assessment.

Four specific objectives were focused on:

- 1) To establish prospective Construct Validity by comparing the PUN-Q to well established parent outcomes measures (parenting stress and perceived self-efficacy), which were hypothesised to be related to parental understanding.
- 2) To examine the test-retest reliability of the PUN-Q (i.e. its stability over time)
- 3) To examine whether or not the PUN-Q is sensitive to changes in parental understanding over time following a comprehensive multi-disciplinary diagnostic assessment.
- 4) To understand more about factors which may be influencing parental understanding by exploring the relationship of the PUN-Q to child emotional, behavioural and social communication difficulties.

It is hoped that the results from this study will add to the previous validation study (Moran et al., submitted) in order to identify the PUN-Q as an effective measure of parental understanding, specific to child neurodisability, which can be used effectively both prior to and following a paediatric diagnostic assessment. The literature review outlined previously established associations between the PUN-Q, parenting stress and perceived self-efficacy with

regards to outcomes for children with neurodisability. To date there has been no systematic examination into the influence that parental understanding of their child's neurodisability may have upon either of these parent-related factors, or the child's emotional, behavioural and social communication outcomes. The PUN-Q is an easy to administer tool, which if shown to be reliable, valid and sensitive to change, can be used to inform clinical practice within neurodisability services. This input will thereby help services to meet the specific needs of parents, who are the advocates of child-related change within any intervention (Ho et al., 1994).

The results from both the previous and current study provide initial evidence to suggest that the PUN-Q is a reliable instrument with which to measure parental understanding within the context of neurodisability. Both within this study and in the previous development and validation phase (Moran et al., submitted), only parents of children with social communication difficulties were recruited. Parental understanding as a concept needs further investigation, however the current study suggests that it is independent, yet related, to two previously established constructs of parenting stress and parental sense of competence (the PSI-SF and PSOC, respectively). Research is now needed to outline the role of parental understanding within previously established models of stress and coping for parents of children with disability (e.g. McConachie, 1994; Hastings, 2002). The next phase of research will therefore be to examine whether the PUN-Q can effectively screen parents' understanding and be sensitive to changing levels of parental understanding within the wider neurodisability service (i.e. not restricted to children with suspected Autism Spectrum Disorder (ASD) symptoms). The ultimate aim is to disseminate the PUN-Q as a valid parent-related outcome measure (PROM) to other services.

The initial study examined construct validity for the newly developed PUN-Q up to two years following a paediatric diagnostic assessment (Moran et al., submitted). The current study

aimed to extend the construct validation by examining associations between the PUN-Q and constructs of parenting stress (PSI-SF) and reported self-efficacy (PSOC), both prior to and immediately following the same diagnostic assessment, within an independent sample of parents. Results supported the a-priori hypotheses showing that prior to the diagnostic assessment, a shortened version of the PUN-Q (the PUN-Q-8) was significantly and negatively correlated with parenting stress and positively correlated with self-reported parenting self-efficacy. The results from this study support findings both the initial PUN-Q validation (Moran et al., submitted) and previous investigations which have shown associations between parental cognitions regarding disability (e.g. appraisals, beliefs and attribution of behaviour), parenting stress and self-efficacy beliefs (see Hassall & Rose, 2005, Jones & Prinz, 2005).

No relationship was shown between the post-assessment PUN-Q-8 or PUN-Q-13 total scores and either parenting stress or parenting self-efficacy beliefs. This result was contrary to the study's a-priori hypotheses and did not support the significant post-assessment associations shown between the same measures in Moran and colleagues' study (submitted). This unexpected result will be further explored, however it is potentially reflective of delayed cognitive change following clinical assessments, which has been previously indicated using parent-professional concordance ratings for children's cognitive abilities (Glaun et al., 1998).

The second aim for this thesis was to examine the test-retest reliability of the PUN-Q-8, which was a novel aspect of this thesis. Comparisons between the two pre-assessment measurements of the PUN-Q-8 (Times 1 and 2) showed no significant differences. This result provides evidence to suggest that the PUN-Q-8 retains stability over a non-interventionist time period (average length of time=6.90 weeks), using a within-subjects design (i.e. the measure is completed by the same group of stable participants).



Also novel to this thesis was the examination of the PUN-Q-8's sensitivity to change; results suggested that the PUN-Q-8 was sensitive to measuring changing levels of parental understanding of their child's neurodisability over time. Time 1 pre-assessment PUN-Q-8 total scores were compared to Time 3 PUN-Q-8 total scores. Results supported the hypotheses by showing a significant difference, with higher levels of parental understanding at Time 3, following the completion of the diagnostic assessment. Post-hoc comparison analyses were conducted to explore the effects of the diagnostic assessment on parenting stress levels and self-efficacy ratings, by comparing Time 1 and Time 3 scores. In these preliminary results no significant increases were shown for either measure. Whilst these post-hoc analyses did not follow any specific hypotheses, they are contrary to other published studies which have shown significant changes to both of the parenting stress and parenting self-efficacy constructs following clinical parenting interventions (e.g. Gardner, Burton, & Klimes, 2006; Plant & Sanders, 2009).

Finally, no significant association was shown between the Time 1 PUN-Q-8 total score (i.e. parental understanding) and levels of children's total difficulties (measured by the SDQ). This result contrasted to the significant associations shown at Time 1 between children's total difficulties (SDQ) and total scores for both parenting self-efficacy and parenting stress. Further analyses compared Time 1 PUN-Q-8 total scores for higher and lower levels of children's social communication difficulties. No significant between-group differences were shown. Comparison analyses showed similar non-significant results for total scores of both parenting self-efficacy and parenting stress. This set of analyses was explorative in nature and therefore did not have stipulated a-prior hypotheses. These results suggest differences to the working of the PUN-Q-8 in comparison to the measures of parenting stress and parenting self-efficacy, when related to measures of child emotional and behavioural difficulties.

## 4.2 Discussion of findings

### 4.2.1 Research Question 1: Pre-Assessment construct validation of the PUN-Q.

No previous study has investigated the construct validity of the PUN-Q prior to clinical intervention. As predicted a-priori, the Time 1 PUN-Q-8 total score was significantly associated with total scores for both parenting stress and parental self-efficacy. Whilst no direct comparison with regards to parental understanding in the context of neurodisability can be drawn from the literature, these results support previous empirical research and theoretical models regarding parental cognitions. These previous studies identify the role of parental cognitions in making parents vulnerable to experiencing greater stress levels and lower levels of parenting self-efficacy beliefs, within the context of child disability (e.g. Human & Teglassi, 1993; McConachie, 1994; Hastings et al., 2002; Trute et al., 2007).

Significant negative correlations were shown between the PUN-Q-8 total score and both the '*parenting distress*' and '*parent-child dysfunction*' subscales of the PSI-SF; the '*parent-child dysfunction*' subscale measures parents' expectations and the satisfaction that they gain through interactions with their child. These results support the hypotheses, suggesting that higher levels of parental understanding are associated with lower levels of parental distress and a greater ability for parents to foster positive interactions with their child, and vice versa.

The negative association between parental understanding of their child's neurodisability and '*parent-child dysfunction*' supports previous studies which have shown that parents of children with complex healthcare needs, or ASD, can find it harder to establish realistic expectations for their child (e.g. Cunningham & Davis, 1985; Mercer et al., 2006). These difficulties can cause parents to misattribute their child's behaviour to non-compliance (i.e. being a '*difficult child*'), and thereby encourage them to utilise harsher parenting strategies (e.g. Lecavalier et al., 2006). Whilst the current results are non-directional, they can be used

to hypothesise that higher levels of parental understanding may influence more realistic expectations for their children. Previous studies have highlighted the difficulties faced by parents of children with ASD in fostering positive interactions with their children (e.g. Johnson & Myers, 2007). Consequently, these results indicate that it would be worthwhile to examine whether clinical interventions, which focus on parental understanding, can aid parent-child interactions; this is unfortunately beyond the scope of the current study.

Contrary to hypotheses, no significant association was shown between the total PUN-Q-8 score and the PSI-SF *'difficult child'* subscale. This suggests that within this sample, higher levels of parental understanding were not directly related to difficulties faced by parents in managing their child's behaviour or gaining their child's cooperation. This result is somewhat surprising, as previous studies have suggested that parents' emotional reactions, which can be a consequence of increased stress, are related to the severity of children's behavioural difficulties, due to a negative reinforcement loop and the adoption of less effective parenting strategies (e.g. Hastings, 2002). Taken together, these results suggest that prior to clinical intervention, higher levels of parental understanding within the context of neurodisability are associated with reduced parental distress levels, but do not directly relate to parents feeling able to cope with their child's difficulties.

As predicted, the Time 1 total PUN-Q-8 score was positively associated with both the PSOC *'parenting satisfaction'* and the *'parental efficacy'* subscales. The latter correlation contrasts to the previous null finding with regards to the PSI-SF *'difficult child'* subscale. Whilst these subscales (i.e. *'parental efficacy'* and *'difficult child'*) were moderately correlated to one-another ( $r=.60$ ), the size of the correlation ( $r<.90$ ) suggests that they are separate constructs (Field, 2009) and thereby tap into different aspects of managing a child's difficulties (i.e. parents' confidence in their practical application of strategies, versus their feelings of being able to cope). Alternatively, these results could support weaknesses to the *'difficult child'*

subscale which have been identified in a previous study (Haskett et al., 2006). The authors' validation of the PSI-SF provided evidence in favour of a two factor model for the PSI-SF, which combined the '*difficult child*' and '*child-parent dysfunction*' subscales into a single '*child-rearing stress*' subscale (Haskett et al., 2006).

The associations between the different aspects of parental understanding with parenting stress and self-efficacy beliefs are clinically important to identify, as increased parenting stress has been related to decline in parents' mental health. Previous studies have demonstrated associations between higher stress levels and maternal depression, in addition to PTSD for parents of children with ASD (Chilcoat & Breslau, 1997, Baylot-Casey et al., 2012). Consequently, parental understanding may be a factor that could hypothetically increase or decrease parental resilience or vulnerability to stress, and thereby influence the development of mental health difficulties. The association between parenting stress and mental health is important to consider with regards to child outcomes, for example, it has been shown to influence the number of appointments that parents attend with their child (Mowery, 2011).

The positive associations shown within this thesis between the PUN-Q-8 and PSOC subscales suggest that higher levels of parental understanding with regards their child's neurodisability is related to parents' enhanced self-efficacy within their role, in addition to higher levels of parenting satisfaction. This result supports a previous study which showed that parental understanding was associated with both parental self-efficacy beliefs and life satisfaction for mothers of children with Autism (Tunali & Power, 2002). These associations are important for clinical services to monitor, especially when taking into account previously documented relationships between perceived parenting self-efficacy and parenting stress levels (e.g. Kuhn & Carter, 2006, Dellve et al., 2006).

Examination into the relationships between the Time 1 PUN-Q-8 '*insightful understanding*' and '*application of understanding*' factors with the PSI-SF and PSOC subscales showed that the '*insightful understanding*' factor (e.g. understanding how their child 'sees' the world) was related only to parenting stress (as measured by the PSI-SF total score) and the '*parent-child dysfunction*' subscale. '*Insightful understanding*' was not associated with the PSOC total score or either of its two subscales. Contrastingly, the PUN-Q '*application of understanding*' was related to the total PSOC score and both its '*parental efficacy*' and '*parenting satisfaction*' subscales. These results indicate that parents' practical awareness of their child (e.g. how to adjust themselves as parents, and knowing what to expect of their child), augurs towards higher self-efficacy beliefs. This somewhat supports previous studies, which have shown that higher parenting self-efficacy enables parents to adjust their parenting strategies even during demanding or stressful parenting experiences (Giallo et al., 2008), i.e. that which could be experienced by parents of children with ASD. Neither of the PUN-Q subscales were related to parental difficulties with their child's behaviour, as measured by the PSI-SF '*difficult child*' subscale. Interestingly, the PSOC pre-assessment '*parental efficacy*' and '*parenting satisfaction*' subscales were both significantly associated with all of the concurrent parental stress subscales.

The results have shown overall evidence for construct validity. The non-significant associations between the PUN-Q and the PSI-SF and PSOC measures are interesting as they provide evidence to suggest that the PUN-Q is a separate measure. Further, the differences in the associations shown between the PUN-Q factors and these measures suggest that different facets of understanding may augur towards risk or resilience for different parenting outcomes. Both of the PSI-SF and PSOC tools contrast to the PUN-Q, as neither were developed specifically for parents of children with neurodisability. It seems therefore that the PUN-Q measure is tapping into a separate area of parent functioning, i.e. more specifically related to a child's difficulties within the context of neurodisability, in comparison to the parenting

factors measured by the PSOC and PSI-SF. This suggests that clinical understanding into the effects of parenting a child with special health-care needs could be enhanced through the utilising the PUN-Q.

#### **4.2.2 Research Question 2: Test-retest reliability of the PUN-Q.**

This study administered the PUN-Q-8 on two occasions before the commencement of the diagnostic assessment, therefore allowing an examination of the PUN-Q's prospective validity using a within-subjects longitudinal design. This is an important step within a new tool's development in order to ascertain whether the scale measures its intended latent construct in a consistent manner, over two stable (i.e. non-intervention) time points (Guyaat et al., 1992). This is the first time that the test-retest reliability of PUN-Q has been examined. As aforementioned, results showed no significant differences shown between total PUN-Q-8 scores measured pre-assessment at Times 1 and 2; i.e. the PUN-Q-8 showed stability over time. This suggests that without any clinical input, parents' understanding within the context of their child's neurodisability symptoms does not vary significantly over time. This stability over time increases the likelihood that any post-assessment increase to parental understanding may be related to the receipt of clinical services. These analyses represent an important additional phase to the validation of the PUN-Q, to provide further evidence that it can be used reliably within clinical settings.

#### **4.2.3 Research Question 3: Sensitivity of the PUN-Q to detect change.**

The stability shown for the PUN-Q-8 over the two pre-assessment time points can help to indicate whether any post-assessment changes are attributable to subsequent clinical input (Guyaat et al., 1987). In addition to assessing the stability of the PUN-Q over time, further investigation of a scale's usefulness is determined by its ability to detect small but important clinical changes over time (Wyrwich et al., 2005). Results supported a-priori hypotheses by

showing that total PUN-Q-8 scores were significantly higher at Time 3 (post-assessment) in comparison to Time 1 (pre-assessment). Significant post-assessment increases were also shown for both of the PUN-Q-8 subscales. Effect sizes indicated that these changes were between small to moderate in magnitude (Cohen, 1992). The contrast between the aforementioned stability of the PUN-Q-8 scores shown between the pre-assessment time points, and the significantly higher post-assessment scores, suggests that the PUN-Q is sensitive to detect small to moderate changes to parental understanding, which may occur following a comprehensive MDT diagnostic assessment.

These results support outcomes from a recent short-term longitudinal study, which showed that MDT assessments were significantly able to enhance parents' understanding of their child's difficulties (Mittal et al., 2014). This study was however conducted with parents of children with mild Learning Disabilities and is therefore not directly comparable to the current study; its findings were additionally limited by the lack of a systematic measure with which to measure parental understanding - analyses were based on single-item questions. The current validation of the PUN-Q therefore adds to the literature by enabling a more thorough examination of the impact that MDT assessments may have for parental understanding.

A further preliminary investigation aimed to examine whether the increased PUN-Q-8 scores could be attributed to the diagnostic assessment. The significance of the association between the total PUN-Q-8 scores at Times 1 and 3 was shown to diminish when accounting for the variance explained by parents' post-assessment understanding (e.g. *'getting a diagnosis confirmed what I already knew about my child'*). These 'post-assessment' PUN-Q items tap into aspects of parental understanding which are directly related to the clinical assessment. Importantly, parent and child factors (as measured by the PSOC, PSI-SF and SDQ) did not significantly contribute to the variance within this model. These results tentatively suggest that the diagnostic assessment may have influenced an increase to parental understanding. It

is not however possible to ascertain whether these changes were a consequence of the current clinical input, or any previous interventions that the parents may have experienced.

Interestingly, contrary to a-priori hypotheses, no significant associations were shown between post-assessment Time 3 total scores for the PUN-Q, PSOC or PSI-SF. These results contrast to the significant post-assessment associations demonstrated by Moran and colleagues (submitted). These results most likely reflect the small Time 3 sample and therefore need replication before any firm conclusions can be drawn. However, taking limitations into account, these results raise an interesting question regarding the optimum time-point at which to administer the PUN-Q, or other parental perception measures. Moran and colleague's (submitted) study was conducted up to two years post-assessment, whilst the current study administered the Time 3 measures soon after completion of the diagnostic assessment; parents answered Time 3 questionnaires on average 27.82 days ( $sd=20.28$ ) after the end of the assessment process. Consequently, the results from the two studies are not directly comparable: for example, the majority of the parents within the initial study received their final clinical report and recommendations before taking part in the study, yet this was not true for any of the parents within the current study. Further, Glaun and colleagues' (1998) study showed that parents' understanding of their child's cognitive abilities increased significantly six months following clinical intervention, in direct comparison to their understanding immediately after the intervention. Subsequently, a further follow-up is needed in order to investigate this further. This is beyond the scope of the current study, but will be included within the final analyses of the study to be completed in September 2014.

In contrast to the increased post-assessment PUN-Q-8 total score, no significant increases were shown for either the PSOC and PSI-SF total scores. Previous studies have shown both of these measures to be sensitive to change following clinical intervention (e.g. Gardner et al., 2006; Plant & Sanders, 2006). Whilst these are secondary outcomes for the current study and



should be interpreted with caution, these results may again reflect a measurement issue regarding the small sample and timing of the final follow-up. The measurement of parental opinions soon after the assessment may have allowed time for parents to make cognitive changes, but would not have given them or anyone within their wider support network (e.g. school teachers and local paediatricians), sufficient time to incorporate any of the practical treatment recommendations into their child's care.

#### **4.2.4 Research Question 4: Exploring the relationship between the PUN-Q and child difficulties.**

This study provided an initial examination into associations between the PUN-Q and well-established measures of child emotional, behavioural and social communication difficulties (SDQ and SCQ). Interestingly, the non-significant association shown between the Time 1 total PUN-Q-8 score and total child difficulties (as measured by the SDQ: Goodman, 1997), contrasted to the significant associations identified between the total child difficulties and both the PSOC and PSI-SF total scores. This result, which may be influenced by the small sample size (n=16) suggests that parental understanding does not directly correlate with child-related emotional and behavioural difficulties. Whilst there were no stipulated a-priori hypotheses for this research question, this is contrary to previous studies that have shown links between parental cognitions and child-related outcomes (see review by Hassall & Rose, 2005). It is possible that a non-linear relationship may exist between parental understanding and child difficulties, for example, this relationship may be mediated by parent factors such as parenting stress or self-efficacy. Exploration into the potential mechanisms underlying this possible model is beyond the scope of the current study, yet important to consider for future research.

### **4.3 Limitations**

This study has presented preliminary analyses for an ongoing longitudinal validation of the recently developed PUN-Q measure. When interpreting these results a number of limitations must be taken into account. As a consequence of these at times substantial limitations, any generalisations or interpretations should be made with caution.

#### **4.3.1 Setting.**

This study recruited parents from one clinic within a Tier-Four specialist Neurodisability service. The children who are referred and accepted to this clinic have complex symptom presentations which require further expertise. This was shown by the high levels of parent reported difficulties, as measured by the SDQ. Twelve children (n=16) scored within the ‘abnormally high range’ (Goodman, 1997); nine out of the fifteen children for whom data were available scored within the risk threshold for ASD (as scored by the SCQ). Further, the average age of children referred to the clinic was 9.73 years, which is higher than the estimated national average for diagnosing ASD (four to five years old: Baird et al., 2006). The sample of parents included in this study may therefore not be representative of parents attending local Tier-2 services; further work is needed to determine the generalisability of these results.

A wide age range of children were included within this study (3.25 to 16.67 years old), reflecting the age of children accepted to the clinic (zero to eighteen years old). Age differences may indicate that the participating parents were at different stages of the diagnostic process, with parents of older children having experienced potentially longer periods of diagnostic uncertainty (e.g. Howlin & Asgharian, 1999; Mansell & Morris, 2004). However, age was not shown to significantly impact parent-related measures within this sample.

Children referred to the clinic often have comorbid disorders; within the past two years, fourteen children included within this study had a neurodevelopmental diagnosis, whilst fifteen had received a medical diagnosis. Consequently, the parents of children invited to participate in this research may have been in contact with a large number of health-care professionals. Many of the children were referred due to atypical presentations, a variety of comorbid disorders, or disputed findings from previous ASD assessments or diagnoses. These referrals therefore reflect a need in either the parents or the local paediatricians to better understand the child's presenting symptoms. Therefore, if effective, the clinical input provided by this service should enhance parental understanding for all parents, regardless of symptom presentation or diagnostic status. Indeed, previous diagnoses were not shown to significantly affect total scores for any of the measures.

#### **4.3.2 Sampling Issues - General.**

Parents varied with regards to the number of appointments that they attended for the assessment and the time in-between these appointments. Time factors were influenced by both family requirements and appointment availability. Staffing difficulties caused some inconsistencies to the arrangement of appointments. For example, at the start of the recruitment process, the clinic's secretary was off work due to sickness, leading to some last-minute appointments being booked in. Subsequently, the length of the assessment process differed substantially between families (range of 7.86 to 20 weeks). The short-notice appointment bookings, led to difficulties collecting both of the pre-assessment time points. This led to delays in the recruitment process at the start of the study, which had a knock-on effect for the number of post-assessment (i.e. Time 3) questionnaires that could be completed within the study's timeframe.

During the recruitment process, some parents reported their qualitative experiences of parenting a child with complex healthcare difficulties. Many parents described being overwhelmed by multiple appointments for their child across different clinics and the high volume of letters which they felt that they needed to respond to. A few parents disclosed the stress that they were experiencing within the diagnostic process. One parent stated that they had been waiting years to gain a referral for the clinic, which they described as their ‘*panacea*’; a number of different services had been previously unable to provide a conclusive diagnostic opinion for their 11 year old child. A further parent stated: “*As time passes by we generally feel that no medical professionals can help... We have to accept our son for who he is and learn to live with this condition*”. As a consequence of this qualitative feedback, a minimum age limit was established so that parents of referred children who were younger than five years old were not invited to participate. The additional stress in coming to terms with their child’s difficulties (see Dale, 1996) may have caused extra distress for parents of younger children, or made it harder for them to commit to the study’s demands and strict time-frame.

The strict timeframe of the study created further difficulties for parents. This was also found to be the case in the previous phase of this study (Moran et al., submitted) – eight parents returned their questionnaires after the completion of the study. There was some confusion with different research projects - some of the parents had been simultaneously invited to participate in other research projects that were being conducted within different, unrelated clinics in the hospital. This led to some parents returning the questionnaires to the wrong department; on one occasion the confusion between studies led to a parent being informed not to return the questionnaires for the current study.

The difficulties experienced by our parents made an ethically sensitive recruitment procedure essential. I contacted parents over the phone and if they stated a wish to participate, I gained

verbal permission to contact them again; I did not repeatedly call parents. I reiterated information about confidentiality, which guaranteed that clinicians would not be aware of who was participating, nor receive any information given by parents for the purpose of the study. Importantly, parents were reminded that decisions regarding participation would not affect their child's clinical care. As a consequence of this study being non-interventionist, I reminded parents that clinicians would not be informed of any qualitative information disclosed regarding difficulties. I encouraged these parents to seek support from their clinicians.

Perhaps due partially to these recruitment difficulties, the current study achieved a relatively small sample size ( $n=37$ ). In order to be statistically powerful to detect medium-sized effects, the study needed 32 participants at each of the time points. Analyses were therefore under-powered at Times 2 and 3 ( $n=26$ ,  $n=11$ , respectively). This study utilised a short form of the PUN-Q at the two pre-assessment time points (referred to as the PUN-Q-8 within this study). The PUN-Q was developed with parents following completion of the diagnostic assessment. The current study was the first time that the PUN-Q has been used as a pre-assessment screening measure. Consequently, the five 'post-assessment' items were omitted from the PUN-Q pre-assessment time points in order to prevent any confusion that they may have caused. The psychometric properties of the PUN-Q-8 need to be further examined. However, the small sample size did not allow factor analysis to be conducted within this study for the use of the PUN-Q-8 both pre and post assessment (Tabachnick & Fidell, 2007); this will be conducted within the final analyses. All results from this study should take this substantial limitation into account.

In order to overcome the small sample size, all analyses were conducted with bias corrected and accelerated bootstrapped confidence intervals. This non-parametric method is favoured for small sample sizes ( $n \leq 30$ ) (Zhu, 1997), and has been shown to provide reliable results as

long as  $n \geq 8$  (Zhu, 1997). This method increases statistical power for smaller sample sizes as it does not assume normality of data, thereby decreasing the probability for Type I and Type II errors (MacKinnon et al., 2002). Bootstrapping can however provide misleading results if the sample is not representative of the overall population (Zhu, 1997), therefore a larger sample would have been preferable with regards specifically to the Time 3 analyses. Analyses were repeated and the results from the bootstrapping confidence intervals were upheld, suggesting that these results reflect the true direction of the stated associations (Davidson & McKinnon, 2001).

#### **4.3.3 Sampling Issues – Parents.**

The majority of parents who agreed to participate were Caucasian mothers and educated to degree level or above. Twenty-two parents were educated to graduate or postgraduate level. Health literacy is in part determined by an individual's cognitive abilities (Nutbeam, 1998), therefore those adults who have completed fewer years of education may show lower cognitive skills and subsequently have lower levels of health literacy. Whilst level of education was not related to any of the parent-related measures within this sample, in some respects this was a self-selecting sample as parents chose whether or not to opt into the study (50% of the total invited parents agreed to participate). It was not however possible to ascertain within this study whether any differences with regards to parental understanding existed between the parents who chose to participate or not. It could be hypothesised that parents who chose to participate were better informed, had a better pre-existing understanding of their child or a higher level of perceived self-efficacy, in comparison to parents who declined participation. Further, taking into account the high complexity of child difficulties, some of the questions which focused on personally sensitive topics may have been too difficult for parents to answer.

The setting of this study may also augur towards a self-selecting sample. Referrals to Tier-Four clinics require parents to persevere and pressurise services; some parents do not receive support initially from local teams, due to lack of funding or parent-professional disagreements regarding a previous diagnostic assessment. Consequently, parents may have needed to be proactive and feel empowered in order to disagree with their local health-care professionals and seek out a specialist neurodisability service. Accordingly, parents with lower levels of education, parental understanding or those experiencing higher stress levels may have found it harder to access this clinic.

Levels of parental stress may have also affected sampling. This study was based within the assessment period of ASD, during which time parents typically experience higher stress levels (Mansell & Morris, 2004). Higher stress levels are associated with decreased parental access to health-care services (Mowery, 2011), consequently, such parents may have also been less likely to participate in healthcare research. Parents who were experiencing higher stress levels may not have been able to accommodate the time demands necessary for participation in the study. However, taking these factors into consideration, this study achieved a greater acceptance rate in comparison to the previous phase of the study conducted in 2011.

#### **4.3.4 Sampling Issues – Children.**

In order to ensure a homogeneous sample, only children who were referred to the clinic with questions regarding social communication were invited to participate in this study. One reason for this group being chosen is that queries regarding ASD account for the majority of new referrals to the clinic (approximately 80%). The PUN-Q was developed by interviewing parents of children diagnosed with a range of neurodevelopmental disorders. The validation of the PUN-Q has however only occurred on one sub-sample, consequently the validity of the PUN-Q for non-ASD groups needs to be determined in future research.

Children may have differed with regards to IQ levels, which were not measured within the current study. It is therefore not possible to determine which aspects of the neurodisability specifically affected parental understanding – whether it was affected by the children’s emotional, behavioural and social communication difficulties, or as a consequence of IQ; seventy-five percent of children with ASD have intellectual disability and delays in development of play skills and self-care (Smith, 1999). Gender differences were apparent within this sample, only four children were male. This reflects other studies; a 3:1 male-female ratio is characteristic of autistic samples (Rutter & Garmezzy, 1983).

#### **4.3.5 Measurement Issues.**

Most of the data collection relied on postal surveys; a few Time 2 questionnaires were completed over the phone due to time constraints. As a consequence of this methodology, it is difficult to ascertain whether the questions were answered accurately or honestly. The questionnaire battery did not include any sham questions or questions which were intentionally contrasting that could have directly tested this. However, other studies have shown a higher level of honesty for postal surveys in comparison to either telephone or face-to-face questioning, due to a higher level of perceived confidentiality; (Bernard et al., 2007; Denscombe, 2007).

As part of the study’s procedure, parents were asked to fill out two sets of questionnaires before they attended their first appointment. The first set of questionnaires was received before parents had been contacted by the clinical team who did not send out acknowledgement letters to parents. This caused confusion for some parents, whilst other parents may have chosen not to participate as they were not yet invested within the clinical process.



Due to time constraints, this study has not assessed parents following the receipt of the full clinical report; this will be conducted within the final analyses for the study. As aforementioned, this may have affected parental understanding, however no new information is included within the final report and parents received intermediate short-reports during the assessment, and a full feedback session with the consultant member of the MDT; these reports were also disseminated to local consultants and school staff. It is also possible that the full report, which is very long, may not be accessible for all parents. Subsequently, we thought it justified to examine treatment effects at 'Time 3'; as have been reported within this thesis.

The measurement of ethnicity was limited as parents were not provided with a guide for standard ethnicity groupings, and as a consequence many parents responded with 'British' as their ethnicity, or chose not to respond to this question.

#### **4.4 Strengths**

This study has incorporated a longitudinal design across three time points, in order to assess the reliability and validity of the PUN-Q measure; following guidelines for good practice set out by Guyaatt and colleagues (1987). This design also allowed an examination into the effectiveness of the Tier-Four assessment service in enhancing levels of parental understanding and self-efficacy ratings, in addition to decreasing levels of parenting stress.

The PUN-Q is a short and easily administered self-report scale for parents. It has been shown to have satisfactory psychometric properties across two similar yet independent samples. The PUN-Q is a novel and potentially useful tool, which both the current and previous studies indicate may tap into a hypothetical construct of parental understanding, within the context of child neurodisability. This measure can be used both to ensure optimal outcomes for children

and their parents, as well as to audit services' ability to effectively communicate with parents regarding information about their child's diagnosis, support needs and treatment.

This study contributes towards the development of the PUN-Q measure as the first PROM to focus directly and systematically on measuring parent's understanding of their child's neurodisability symptoms. This is a concept which has been identified within the literature as important to assess directly (e.g. Glaun et al., 1998; Tunali & Power, 2002). Previous studies have only crudely measured, for example, they have used one item within a larger questionnaire battery in order to examine whether parents of children with ASD 'understand' their child (Tunali & Power, 2002; Mittal et al., 2014). It is hoped that the findings from this study can facilitate further comprehensive and systematic examinations of parental understanding both in future research projects, in addition to routine clinical practice.

This study recruited only those children had been referred to the Tier-Four service for questions regarding potential ASD. Homogeneity of diagnosis helped to minimise the effect that differential disorders may have had on parental understanding and the additional variance that this may have accounted for (Prince, 2003). This is especially important within the context of Tier-Four services who accept children with highly complex and varying symptom presentations which may differentially affect parental understanding. Whilst it was not possible to fully match parents, homogeneity of diagnosis ensured that all parents were coping with a similar genre of symptoms in their children.

The Tier-Four setting ensured that each child was assessed by the same clinical multi-disciplinary team, which is comprised of specialists who are experts at assessing and diagnosing ASD in children with complex presentations. The expertise of the team includes their ability to effectively explain the outcome of the assessment to parents. This is important as it reduces the variability in the quality of the assessment that the parents received, and

therefore increases the probability that any differences in understanding are due to idiosyncratic parent-related factors, rather than as a consequence of the quality of services received.

#### **4.5 Clinical Implications**

This study has shown the newly developed PUN-Q to have good to strong internal reliability for both the shortened pre-assessment scale and the longer post-assessment scale. In practice, this means that the PUN-Q can fulfil a number of clinical purposes. For example, it can be used as a short and reliable self-report screening measure of parental understanding of their child's neurodisability, both prior to and following a clinical assessment. The addition of the PUN-Q to the literature will allow services to conduct systematic evaluations into the effectiveness of their interventions for enhancing parental understanding. This will build upon previous investigations which have crudely shown MDT assessments of ASD to enhance parental understanding (Mittal et al., 2014).

As part of the clinical governance process, service providers are required to seek out appropriate measurement tools, which are sensitive and specific to different aspects of health-care provision (Fayed et al., 2012). Recent government policies, such as '*Making Mental Health Matter More*' (Department of Health, 2014) and '*Liberating the NHS: No decision about me without me*' (Department of Health, 2012) outline the importance of service-user involvement and shared decision making in the provision of healthcare. Within these guidelines, service-users and their representatives are afforded more control over their own care: before, during and after the diagnostic processes. These policies build on well-established ideas within the disability literature, such as 'parents as partners' or 'Family Centred Care' models (e.g. Squires et al., 1996; Law et al., 2003). The newly developed and validated PUN-Q can be used by services to help implement these ideas, by allowing the

systematic screening and monitoring of parental understanding levels (both pre-and-post clinical intervention). This process may help to identify parents' support needs and thereby enable them to make informed decisions and advocate effectively for their child's healthcare needs.

The current study therefore builds on the understanding that parents should be seen as partners and essential within the treatment of their child. The validation of this non-functionally focused PROM (see Fayed et al., 2012), which focuses instead on parents' understanding of their child and the impact of their child's difficulties, within the context of neurodisability, recognises the importance of parents within the diagnostic process. Further, the PUN-Q examines the specific, individual needs of the parent. This focus on the parents' role within the diagnostic assessment process will ensure the fulfilment of 'patient centred care', as stipulated by NICE guidelines for the assessment of ASD (NICE, 2011), and help neurodisability services to tailor their practices to meet parents' individual needs as carers for their children.

With regards to the Tier-Four service in which this study took place, these results indicate that the diagnostic assessment may help to enhance parental understanding of their child within the context of neurodisability. This has been shown to be important due to links between parents' cognitions of their child, and subsequent child outcomes, for example with regards to parents' attributions of their child's behaviour (Chavira et al., 2000), or their estimation of their child's abilities (Geiger et al., 2002).

#### **4.6 Theoretical Implications**

The concept of parental understanding, which has been explored to a lesser extent in previous studies (e.g. by studies using single-item questions: Tunali & Power, 2002) has been further

developed by this thesis. The results from this study suggest that this cognitive aspect of parenting is important to consider specifically within the context of neurodisability clinical care. Importantly, parental understanding has been shown to be fluid over time, such that it can be enhanced by interventions, and as a hypothetical construct it is related, yet independent to other well-established parenting constructs (e.g. parenting stress and perceived self-efficacy); this has been shown by correlations which are substantially lower than  $r=.90$ , which is the threshold for multicollinearity (Field, 2009). Further investigation is however needed regarding the manner in which parental understanding fits together with these established constructs, to enable an overall understanding of how parents cope when caring for a child with neurodisability.

Different models within the literature have previously established theoretical links between parents' cognitive styles, their experience of stress and outcomes for their child. For example, Hastings (2002) proposes that parental cognitions (e.g. beliefs, self-efficacy) mediate or explain the relationship between parental stress and children's outcomes. This model however proposes a linear relationship between parents' cognitions and both child and parent-related outcomes. The differences shown between the post-assessment results for the current study in comparison to Moran and colleague's (submitted) study, suggest that these relationships are not necessarily static and may change pre-and-post intervention, and then continue to change some time after receiving the intervention; supporting previous empirical research (Glaun et al., 1998). Such changes may reflect unmeasured parental cognitive process, or they could potentially be associated with a child's development. Hasting's (2002) linear model of parental cognition and outcomes is perhaps overly simplistic and a more comprehensive model of parenting, within the context of neurodisability, is required in order to assess whether cognitive change lead to behavioural change and whether this is stable or fluid over time.

A further well-cited model proposed by McConachie (1994) linked parental cognitive coping strategies (e.g. attitudes towards disability and beliefs regarding causation) to child, parent and family based outcomes (e.g. adaptation) for children with disabilities. Whilst explaining this model, McConachie (1994) suggests that parents who are able to utilise problem-focused coping strategies (e.g. planning) show better longer-term outcomes.

Hypothetically, parental understanding may fit within McConachie's (1994) model as a moderator in the association between parenting stress, attainment of coping strategies and child or parent related outcomes. For example, parents with higher levels of understanding regarding their child may be better able to incorporate and utilise adaptive coping strategies, or target behavioural interventions in a more developmentally accurate manner. This postulation is supported somewhat by a recent intervention study, which showed that providing individualised information and support for parents of children with ASD was more effective at lowering parenting stress and increasing perceived self-efficacy, in comparison to a generalised video-based intervention (Keen, Couzens, Muspratt, & Rodger, 2010).

Preliminary analyses from the current study have suggested that a diagnostic assessment may help to enhance parental understanding of their child's neurodisability symptoms. Interestingly, no effect of the assessment was shown for either parenting stress or perceived parental self-efficacy. This could reflect measurement difficulties (as discussed previously), however it could also indicate a potential role for parental understanding as a moderator in reducing stress and enhancing self-efficacy. It is beyond the scope of the current study to explore the mechanisms behind this change. Further investigation is therefore needed to explore a more comprehensive model of parenting within the context of neurodisability, taking into account the evidence from this and Moran and colleagues' (submitted) studies regarding the importance of parental understanding.

In support of this hypothesis, Hastings and Beck's (2004) review of the intervention literature shows that Cognitive Behavioural Therapy based interventions are the most effective in reducing stress for mothers of children with intellectual disabilities; i.e. those interventions that incorporate both cognitive and behavioural elements. The authors conclude that further explanation is needed regarding the process of clinical change within these interventions. Clinical measurement of parental understanding could potentially help to explain the mechanisms underlying these interventions and consequently help parents to maintain any positive effects with regards to parental coping and stress. Such maintenance factors are as yet unclear within the ASD intervention literature (see review by Matson, Mahan & Matson, 2009).

#### **4.7 Future Directions**

As aforementioned, this study is part of an ongoing project. The overall study will include a fourth time point, which will assess parental understanding of their child's neurodisability following parents' receipt of the full clinical report (approximately 6 to 8 weeks after the completion of the diagnostic assessment). The inclusion of a fourth time-point will allow further exploration of parental understanding and its associations with parenting stress and perceived self-efficacy. 'Time 4' will not be immediately after the diagnostic assessment and may therefore increase the possibility that parents will have been able to integrate the information provided to them, incorporate behavioural recommendations or cognitive changes, and to see potential improvements within their child. Further, at this time-point the clinical report would have been received by the parents' wider support networks (e.g. local paediatrician and school-staff), which may have facilitated changes to the child's wider environment.

The PUN-Q was developed to be used clinically, however it has only been utilised thus far within research and postal surveying methods. The overall aim of this research is to enable the PUN-Q to be completed by parents as part of routine clinical practice. Additional validation is therefore necessary in order to ascertain whether it can be used within a clinic setting. Further, the PUN-Q has been validated for use within a Tier-Four setting, further validation is needed for its use within Tier-Two and Three services, which provide input for children with less complex symptoms, and at an earlier stage within the diagnostic process.

Factors related to parental understanding (e.g. parenting stress) have been shown in previous studies to be related to poorer mental health outcomes in parents of children with ASD (e.g. Beck et al., 2004; Herring et al., 2006; Baylot Casey et al., 2012). Further research is needed to determine whether parental understanding is related to parental mental health, or involved indirectly within a more complex model in relation to other parenting factors (e.g. stress and self-efficacy).

Further examination is also needed to assess whether the PUN-Q works differently for fathers and mothers; this was beyond the scope and the statistical power of the current study. Certain child factors which are relevant to parenting a child with ASD, have been shown within the literature to have differential effects on mothers or fathers. For example, factors effecting self-efficacy beliefs and stress levels (Hastings & Brown, 2002; Ornstein-Davis, 2008; Herring, 2006). Many of the studies identified within the literature review examined the impact of parenting a child with ASD for mothers only (e.g. Tunali & Power, 2002; Hassall et al, 2005; Kuhn & Carter, 2006; Tomanik et al, 2004). Consequently, future research should focus on recruiting fathers in order to investigate these factors for both parents – this is important in order to understand the wider context in which a child is parented, which may influence child-related outcomes.



The current study collected information on ethnicity - only three parents identified as non-Caucasian (three parents did not provide this information). It was therefore beyond the scope of this research to investigate potential differences between ethnic groups. One limitation of the PUN-Q which was identified within the initial validation stage is that whilst the measure was developed using the views of service users, six of the seven included parents identified their ethnicity as White British. Further investigation is needed to ascertain whether the PUN-Q can be used reliably within different ethnic groups, and whether ethnic groups will differ with regards to parental understanding, or indeed in their access of neurodisability services. Other interventions, for example, school-based adolescent drug and alcohol interventions (e.g. the Keepin' it REAL programme: Marsiglia et al., 2011) have found it necessary to utilise interventions which recognise and incorporate adaptations based on differences between ethnic group differences. Taking this into account, different versions of the PUN-Q may therefore be needed for specific ethnic groups.

Reports have highlighted disparities between various Black and Minority Ethnic and Refugee (BMER) groups and the majority Caucasian population in accessing health services. This was exemplified within the 'Inside Outside' Report, published by the National Institute for Mental-health in Britain (Sashidharan, 2003), which demonstrated that people within BMER groups experience increased social exclusion, influencing poorer health and increased difficulties in accessing the relevant healthcare. Further, BMER communities, in particular the African-Caribbean community, have reported adverse experiences when accessing services through General Practice (Bhugra, Harding & Lippett, 2004), which is regarded as the 'gatekeeper' for specialist health-care treatment. Neurodisability has large overlaps with mental health with regards to associated symptoms and stigma directed towards it, therefore BMER communities may find it harder to access wider neurodisability services in a similar fashion as shown for mental health services. It is important that neurodisability services recognise and monitor such difficulties. Examining differences in parental understanding with

regards to children's neurodisability may help services to tailor interventions, taking into account any ethnic differences, and in this way help to improve access to services for BME communities.

## **4.8 Conclusion**

This study has built upon the previous development and validation of the PUN-Q (Moran et al., submitted), to show that the PUN-Q is a tool which is stable over a test-retest time frame, and can reliably measure parental understanding within the context of neurodisability. Parental understanding is a concept which has been indicated within this study as important for both parent and child-related outcomes - the PUN-Q is the first instrument to enable its' systematic measurement. This study is also the first investigation into the PUN-Q's validity and sensitivity to change both pre-and-post diagnostic assessment. Findings from this study suggest that the PUN-Q is related, yet independent to other well-established parent measures of stress and self-efficacy. Importantly, the current results suggest that parental understanding is fluid over time, such that it can be enhanced by an individualised and comprehensive diagnostic assessment. Further investigation is needed regarding the mechanism underlying this change and the manner in which parental understanding may impact on both parent factors (e.g. stress and self-efficacy) in addition to child emotional, behavioural and social communication outcomes. The PUN-Q has so far only been validated upon parents of children with ASD, further research is needed to evaluate its reliability within the wider context of neurodisability.

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## APPENDICES

### Appendix 1: Ethics Amendment Acceptance Letter



National Research Ethics Service

NRES Committee London - Bloomsbury

HRA NRES Centre Manchester  
Barlow House 3rd Floor  
4 Minshull Street  
Manchester  
M1 3DZ

Tel: 0161 625 7815  
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05 September 2013

Dr Naomi Dale  
Head of Psychology (Neurodisability)  
Great Ormond Street Hospital  
Wolfson Neurodisability Service  
Great Ormond Street Hospital  
Great Ormond Street  
London  
WC1N 3JH

Dear Dr Dale

**Study title:** Development of a measure of parental understanding in child neurodisability services  
**REC reference:** 10/H0713/7  
**Amendment number:** Substantial Amendment 02  
**Amendment date:** 02 October 2009  
**IRAS project ID:** 24835

- The amendment proposes to change the CI from Dr Ian Moran to Dr Naomi Dale.
- It is also proposed to use three additional questionnaires.
- The Sponsor organisation is also changed to Great Ormond Street Hospital.

The above amendment was reviewed by the Sub-Committee in correspondence.

#### Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

#### Approved documents

The documents reviewed and approved at the meeting were:

Document	Version	Date
CI IRAS Declaration	3.5	25 July 2013
Questionnaire: Social Communication Questionnaire (SCQ)	Validated	



Notice of Substantial Amendment (non-CTIMPs)	Amendment 2	02 October 2009
Letter from Dr Ian Moran		04 July 2013
Letter of invitation to participant	3	14 August 2013
Email from Sponsor stating for happy to proceed		12 August 2013
Sponsor IRAS Declaration		15 August 2013
Questionnaire: Client Satisfaction Questionnaire (CSQ)	Validated	
Covering Letter		14 August 2013
Letter from Dr Naomi Dale		04 July 2013
Participant Consent Form	3	04 July 2013
Investigator CV	Naomi Dale	
Questionnaire: Strengths and Difficulties Questionnaire (SDQ)	Validated	
Participant Information Sheet	4	14 August 2013

### Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

### R&D approval

All investigators and research collaborators in the NHS should notify the R&D office for the relevant NHS care organisation of this amendment and check whether it affects R&D approval of the research.

### Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

10/H0713/7:	Please quote this number on all correspondence
-------------	--

Yours sincerely



Signed on behalf of:  
**Professor Faith Gibson**  
**Alternate Vice-Chair**

E-mail: [nrescommittee.london-bloomsbury@nhs.net](mailto:nrescommittee.london-bloomsbury@nhs.net)

## Appendix 2: Parent Recruitment Covering Letter

Dear,

**Title: Development of a measure of parental understanding in child neurodisability**

Thank you for taking the time to read this letter regarding a research project at the [REDACTED]. You have been sent this letter because you have been asked to take part in this research. The sheets attached describe what taking part will involve and what will happen to the information collected about you and your child. It is very important that you read this information carefully before agreeing to take part. In order to give you the opportunity to ask any questions that you may have about this, a member of our clinical team will contact you in the coming week.

If, after reading the attached information sheet and having had the opportunity to ask any questions, you decide that you would like to take part, please read and sign the enclosed Consent Form and then complete the four brief questionnaires, also enclosed.

Please then put these in the enclosed stamped addressed envelope and send them back to us. We estimate that filling in these questionnaires will take about 20-25 minutes in total. Whether you decide to take part in this research or not, the service and clinical care that you and your child receive from [REDACTED] Hospital will not be affected in any way. We are very grateful for parents who are willing to help us to do this research.

Thank you for your attention and time.

Yours sincerely,

Dr. [REDACTED] (Chief Investigator)  
Head of Psychology (Neurodisability)  
Consultant Clinical Psychologist

## **Appendix 3: Parent Information Sheet**

### **Title: Development of a measure of parental understanding in child neurodisability**

You are being invited to take part in a research study. Before deciding whether or not to take part, it is important that you understand why the research is being done and what it will involve. Please take the time to read the following information carefully and to discuss it with others if you wish. The points below tell you the purpose of the study and what we will ask you to do should you decide to take part. Please do ask us if anything is not clear or if you would like more information.

#### **1. What is the purpose of this study?**

The clinic team at the [REDACTED] ( [REDACTED] ) need to know that its work is helpful for the children that they work with and their families. Because so much of the team's work is to assess and diagnose children with developmental concerns and neurodisabilities, the team needs to know that the information that they give to parents about their child has been understood and is useful. We have recently developed a brief questionnaire that parents are able to fill in themselves, which will help us know whether we have communicated information about your child clearly. Now we would like to use this new questionnaire before and after you meet the NAC assessment team with your child. The team is also interested in finding out which other factors are related to parents' understandings about their child and their special needs.

#### **2. Why have I been invited to take part?**

We are inviting all parents whose child has been referred for an assessment at the [REDACTED] Clinic during the study period. We are inviting only those parents where the referral letter mentions concerns about social communication in order to simplify the research by focussing on one referral question only.

#### **3. Do I have to take part?**

No, it's entirely up to you to decide whether to take part. If you do decide to, please sign the Consent Form to say you have agreed to take part. You are free to withdraw at any time, without giving a reason. This will not affect any future care your child may receive.

#### **4. What will happen to me if I choose to take part?**

Taking part will involve filling in some questionnaires about your understanding of your child's development and possible neurodisability, and your experience of being a parent. We will send you a number of questionnaires as soon as your referral has been accepted and you are waiting for your appointment date. We will ask to fill in the questionnaires (this will take about 20 minutes) and please return them immediately in the provided stamped addressed envelope.

Then a few weeks later, just before you attend your appointment, we will ask you to fill in one of the questionnaires again (this takes about 5 minutes) and to return in a second stamped addressed envelope.

After your final appointment at the clinic, we will ask you to complete a number of questionnaires (these will take about 20 minutes). This can be done at the end of your appointment or afterwards and then sent back to us in a stamped addressed envelope.

When you receive the final report, we will ask you to complete one final questionnaire (taking about 5 minutes).

At each time point, the researcher will contact you a week before or after posting out the questionnaires, in order to check that you received them, and to answer any questions that you may have.

You will be asked by the [REDACTED] clinicians to fill in two questionnaires about your child and their behaviour (the Strengths and Difficulties Questionnaire and the Social Communication Questionnaire) before the assessment starts. After your assessment has finished, we would like to use the information obtained from these questionnaires to inform this study.

### **5. What should I do if I want to take part?**

After you have received this Parent Information Sheet, a member of the clinical team will ring you in one week's time to answer any questions and to advise you regarding the Consent process. If you would like to take part, you will need to fill in the Consent Form which is available with the Parent Information Sheet.

You will find the questionnaires enclosed with the Consent Form. Please fill these in and return both the Consent Form and the questionnaires to the researcher in the stamped addressed envelope.

### **6. What are the potential disadvantages to taking part?**

We think that it is unlikely that you will experience any disadvantages from taking part in this research. However, although widely used, some of the questionnaires may contain questions which some participants might find slightly upsetting. If you do decide to take part and find that you have strong feelings after you fill in the questionnaires, you will be offered the opportunity to discuss this with the researcher or the clinician responsible for your child's care.

### **7. What are the potential benefits of taking part?**

There are no direct benefits from taking part, but we hope that the information that we get from this study will help us to improve our service and the way that we work with children and their families.

### **8. What if there is a problem?**

If you have a concern about any aspect of this study, you should ask to speak to the researcher who will do her best to answer your questions. If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints procedure, details of which can be obtained from the Patient Advice Liaison Service [REDACTED] on [REDACTED] or email [pals@](mailto:pals@).

### **9. Who will have access to my child's research records?**

Only the researchers involved in this study. The Sponsor and Regulatory Authorities will require access to data collected during this study in order to monitor and audit the conduct of the study.

We will follow ethical and legal practice in the storage of data, and all information about you and your child will be handled in confidence. All results of this study will be anonymous so your name will not appear on any report of the study. We are following the government's

strict rules about how information like this has to be stored to keep it secure. We may need to keep the research data for up to 25 years.

The only situation where confidentiality will be broken is if we are concerned about your safety or anyone else's. In these exceptional circumstances, we would inform you of our intention before we did this.

**10. What will happen to the results of the study?**

It is hoped that the results of the study will be published in a relevant journal and may be presented at a relevant conference, although participants will not be identified in any way. If you choose to take part and wish to receive a summary of the results, please indicate this on your Consent Form.

**11. Who has reviewed the study?**

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given a favourable opinion by [REDACTED] and the Institute of Child Health Research Ethics Committee.

NB: You may wish to retain this information sheet for reference and contact us with any queries.

Thank you for your time and for considering taking part in the study. If you decide to take part you will be given a copy of the information sheet and a signed Consent Form to keep.

Yours sincerely,

[REDACTED], Dr  
Head of Psychology (Neurodisability)  
Consultant Clinical Psychologist

## Appendix 4: Parent Consent Form

### CONSENT FORM

**Project Title:** Development of a measure of parental understanding in child neurodisability

**Researchers' Names:** Dr. [REDACTED], Dr. [REDACTED], Dr. Lauren Topper, Dr [REDACTED]

**Please tick all the points below in the boxes provided and sign, name and date the form:**

For parent(s) to fill in:

<b>Please tick</b>	
	<b>I confirm that I have read and understood the information sheet for the above study (13/05/2013 V4) and have had the opportunity to ask questions.</b>
	<b>I understand that my participation is voluntary, and that I am free to withdraw at any time without giving any reason and without our legal rights being affected.</b>
	<b>I understand that relevant sections of my medical notes and/or data collected during the study may be looked at by individuals from the sponsor ([REDACTED] Hospital [REDACTED]), from regulatory authorities or from the NHS Trust where it is relevant to my taking part in this research. I give permission for these individuals to have access to my/my child's data and/or records.</b>
	<b>I agree to participate in this study and to complete the study's questionnaires</b>
	<b>I am aware that the data collected as part of this study will be stored in anonymised form for up to 25 years and might be used in future studies</b>

\_\_\_\_\_  
Parent's Name

\_\_\_\_\_  
Parent's Signature

\_\_\_\_\_  
Date

\_\_\_\_\_  
Investigator's Name

\_\_\_\_\_  
Investigator's Signature

\_\_\_\_\_  
Date

NB: This consent form will be stored separately from the responses that you provide.

## **Appendix 5: PUN-Q Scale Development Process**

The following section summarises the initial development stages for the PUN-Q measure (Moran et al., submitted)

### *Item Generation and content analysis*

In order to generate items that could capture parental understanding, individual interviews were conducted with six parents whose child had previously attended the clinic and completed a neurodisability diagnostic assessment. Six interviews were deemed sufficient in order for qualitative coding categories to emerge (Francis et al., 2009). The interviews aimed to develop an understanding of the issues that parents of children with neurodisability symptoms experience when trying to understand their child and his/her difficulties. Parents' contributions towards item generation ensured that items included within the new PUN-Q measure took account of parents' lived experiences of caring for a child with neurodisability symptoms, and their experience of completing the clinical process with their child.

The interviews consisted of open-ended targeted questions covering the main areas of understanding identified within a literature review. Interviews were transcribed following a process outlined by Flick (2009) and summarising content analysis was conducted (Flick, 2009; Weber, 1990), to ensure that the generated items reflected aspects of understanding important to parents. A coding scheme was derived and implemented, followed by two 'reduction' stages (Neuendorf, 2002), which paraphrased and combined areas of the transcript that held similar meanings (Flick, 2009). The content analysis, together with items developed as a result of the literature review, generated 35 potential items, with seven different categories of understanding: diagnosis, difficulties, treatment and recommendations, process of building understanding, prognosis, consequences, and strengths.

The 35 selected items were then rated by a panel of five experts within the clinical team for their relevance and clarity. The team members individually rated each item for its relevance and clarity using a five point Likert scale (Lynn, 1986). This led to the calculation of a Content Validity Index, representing the proportion of experts who endorsed each of the scale's items. This process identified 22 items which were rated as having 'good' content validity. This draft questionnaire was piloted with 12 parents, which is an acceptable sample for piloting a new measure within a rare population (Gillham, 2008). The pilot aimed to

identify ease of use for the draft PUN-Q, and whether any questions were difficult to understand; no difficulties were indicated by any of the parents.

### *Construct Analysis*

The psychometric properties of the draft scale were explored and the data screened following a procedure outlined by Tabachnik and Fidell (2007). Normality of the data was checked using cumulative probability plots and analysis of skewness for each of the 22 items (Field, 2009). This process led to the deletion of 9 items from the scale, due to skewness (Tabachnik and Fidell, 2007) and lack of internal consistency (i.e. item-total correlations lower than  $r=.30$ ) (DeVellis, 2003; Field, 2009). This resulted in a final PUN-Q consisting of 13 items.



## Appendix 6a: The Parental Understanding of Neurodisability Questionnaire - 8

### items

*This is the Pre-assessment version – the PUN-Q-8 administered at Time 1 and Time 2*

This questionnaire contains a variety of statements. Please read each statement carefully and then circle the response that best represents your opinion.

Circle SA if you strongly agree with the statement

Circle A if you agree with the statement

Circle N if you neither agree nor disagree with the statement

Circle D if you disagree with the statement

Circle SD if you strongly disagree with the statement

While you may not find a response that exactly states your feelings, please circle the response that comes closest to describing how you feel.

Circle only one response for each statement and please respond to all statements.

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly Disagree
1) I understand how my child sees the world	SA	A	N	D	SD
2) Most of the time, I understand why my child behaves the way that s/he does	SA	A	N	D	SD
3) There are quite a few aspects of my child's behaviour that don't make sense to me.	SA	A	N	D	SD
4) It isn't clear to me what I can do to help my child.	SA	A	N	D	SD
5) I know how to adjust what I do as a parent to take account of my child's difficulties.	SA	A	N	D	SD
6) I don't really know what is reasonable to expect of my child.	SA	A	N	D	SD
7) I can read or hear about my child's diagnosis, but still struggle to make sense of how it applies to him/her.	SA	A	N	D	SD
8) I could do with someone going through the explanation of my child's difficulties to help me understand it better.	SA	A	N	D	SD

## Appendix 6b: The Parental Understanding of Neurodisability Questionnaire -

### 13 items

*This is the Post-assessment version – the PUN-Q-13 administered at Time 3 only*

#### Instructions:

This questionnaire contains a variety of statements. Please read each statement carefully and then circle the response that best represents your opinion.

Circle SA if you strongly agree with the statement

Circle A if you agree with the statement

Circle N if you neither agree nor disagree with the statement

Circle D if you disagree with the statement

Circle SD if you strongly disagree with the statement

While you may not find a response that exactly states your feelings, please circle the response that comes closest to describing how you feel.

Circle only one response for each statement and please respond to all statements.

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly Disagree
1) I understand how my child sees the world	SA	A	N	D	SD
2) Most of the time, I understand why my child behaves the way that s/he does	SA	A	N	D	SD
3) There are quite a few aspects of my child's behaviour that don't make sense to me.	SA	A	N	D	SD
4) Explanations that I have been given to explain my child's difficulties make a lot of sense to me.	SA	A	N	D	SD
5) It isn't clear to me what I can do to help my child.	SA	A	N	D	SD
6) I know how to adjust what I do as a parent to take account of my child's difficulties.	SA	A	N	D	SD
7) I don't really know what is reasonable to expect of my child.	SA	A	N	D	SD
8) Getting a diagnosis confirmed what I already knew about my child.	SA	A	N	D	SD
9) There is a good fit between the clinical team's understanding of my child and my understanding of him/her.	SA	A	N	D	SD
10) I can read or hear about my child's diagnosis, but still struggle to make sense of how it applies to him/her.	SA	A	N	D	SD
11) I understand the recommendations made for my child.	SA	A	N	D	SD
12) I don't understand how my child's diagnosis fits in with his/her difficulties.	SA	A	N	D	SD
13) I could do with someone going through the explanation of my child's difficulties to help me understand it better.	SA	A	N	D	SD

## Appendix 7: The Parenting Stress Index – Short Form

### Instructions

This questionnaire contains 36 statements. Read each statement carefully. For each statement, please focus on the child you are most concerned about, and circle the response that best represents your opinion.

Circle SA if you strongly agree with the statement

Circle A if you agree with the statement

Circle the NS if you are not sure

Circle the D if you disagree with the statement

Circle the SD if you strongly disagree with the statement

For example if you sometimes enjoy going to the movies, you would circle A in response to the following statement:

I enjoy going to the movies      SA    A          NS      D          SD

While you may not find a response that exactly states your feelings, please circle the response that comes closest to describing how you feel.

YOUR FIRST RESPONSE TO EACH QUESTION SHOULD BE YOUR ANSWER.

Circle only one response for each statement and respond to all statements.

	Strongly agree	Agree	Not sure	Disagree	Strongly disagree
1. I often have the feeling that I cannot handle things very well	SA	A	NS	D	SD
2. I find myself giving up more of my life to meet my children's need than I ever expected	SA	A	NS	D	SD
3. I feel trapped by my responsibility as a parent	SA	A	NS	D	SD
4. Since having this child I have been unable to do new and different things	SA	A	NS	D	SD
5. Since having a child, I feel that I am almost never able to do things that I like to do	SA	A	NS	D	SD
6. I am unhappy with the last purchase of clothing that I made for myself	SA	A	NS	D	SD
7. There are quite a few things that bother me about my life	SA	A	NS	D	SD

	Strongly agree	Agree	Not sure	Disagree	Strongly disagree
8. Having a child has caused more problems than I expected in my relationship with my spouse (or male/female friend)	SA	A	NS	D	SD
9. I feel alone and without friends	SA	A	NS	D	SD
10. When I go to a party, I usually expect not to enjoy myself	SA	A	NS	D	SD
11. I am not as interested in people as I used to be	SA	A	NS	D	SD
12. I don't enjoy things as I used to	SA	A	NS	D	SD
13. My child rarely does things for me that make me feel good	SA	A	NS	D	SD
14. Sometimes I feel my child doesn't like me and doesn't want to be close to me	SA	A	NS	D	SD
15. My child smiles at me much less than I expected	SA	A	NS	D	SD
16. When I do things for my child, I get the feeling that my efforts are not appreciated very much	SA	A	NS	D	SD
17. When playing, my child doesn't often giggle or laugh	SA	A	NS	D	SD
18. My child doesn't seem to learn as quickly as most children	SA	A	NS	D	SD
19. My child doesn't seem to smile as much as most children	SA	A	NS	D	SD
20. My child is not able to do as much as I expected	SA	A	NS	D	SD
21. It takes a long time and it is very hard for my child to get used to new things	SA	A	NS	D	SD

For the next statement choose your responses from the choices "1" - "5" below 22. I feel that I am: 1. Not very good at being a parent 2. A person who has some trouble being a parent 3. An average parent 4. A better than average parent 5. A very good parent	1	2	3	4	5
---	---	---	---	---	---

23. I expected to have closer and warmer feelings for my child then I do and this bothers me	SA	A	NS	D	SD
--	----	---	----	---	----

	Strongly agree	Agree	Not sure	Disagree	Strongly disagree
24. Sometimes my child does things that bother me just to be mean	SA	A	NS	D	SD
25. My child seems to cry or fuss more often than most children	SA	A	NS	D	SD
26. My child generally wakes up in a bad mood	SA	A	NS	D	SD
27. I feel that my child is very moody and easily upset	SA	A	NS	D	SD
28. My child does a few things which bother me a great deal	SA	A	NS	D	SD
29. My child reacts very strongly when something happens that my child doesn't like	SA	A	NS	D	SD
30. My child gets upset easily over the smallest things	SA	A	NS	D	SD
31. My child's sleeping or eating schedule was much harder to establish than I expected	SA	A	NS	D	SD

**Continued on next page**

For the next statement choose your response from the choices "1" to "5" below 32. I have found that getting my child to do something or stop doing something is: 1. Much harder than I expected 2. Somewhat harder than I expected 3. About as hard as I expected 4. Somewhat easier than I expected 5. Much easier than I expected	1	2	3	4	5
For the next statement choose your responses from the choices "10+" to "1-3" 33. Think carefully and count the number of things which your child does that bother you. For example: dawdles, refuses to listen, overactive, cries, interrupts, fights, whines, etc.	10+	8-9	6-7	4-5	1-3

	Strongly agree	Agree	Not sure	Disagree	Strongly disagree
34. There are some things my child does that really bother me a lot	SA	A	NS	D	SD
35. My child turned out to be more of a problem than I had expected	SA	A	NS	D	SD
36. My child makes more demands on me than most children	SA	A	NS	D	SD

## Appendix 8: The Parenting Sense of Competence Scale

Using the 1 to 6 scale, please circle the number on the right that best reflects your feeling about the following statements:

**1 = Strong Agree 2 = Agree 3 = Slightly Agree 4 = Slightly Disagree 5 = Disagree 6 = Strongly Disagree**

1. The problems of taking care of a child are easy to solve once you know how your actions affect your child, and understanding <u>that you have acquired</u> .	<b>1 2 3 4 5 6</b>
2. Even though being a parent could be rewarding, I am frustrated now while my child is at his/her present age.	<b>1 2 3 4 5 6</b>
3. I go to bed the same way I wake up in the morning, feeling I have not accomplished a whole lot.	<b>1 2 3 4 5 6</b>
4. I do not know why it is, but sometimes when I'm supposed to be in control, I feel more like the one being manipulated.	<b>1 2 3 4 5 6</b>
5. My mother/father was better prepared to be a good mother/father than I am.	<b>1 2 3 4 5 6</b>
6. I would make a fine model for a new mother/father to follow in order to learn what she/he would need to know in order to be a good <u>parent</u> .	<b>1 2 3 4 5 6</b>
7. Being a parent is manageable, and any problems are easily solved.	<b>1 2 3 4 5 6</b>
8. A difficult problem in being a parent is not knowing whether you're doing a good job or a bad one.	<b>1 2 3 4 5 6</b>
9. Sometimes I feel like I'm not getting anything done.	<b>1 2 3 4 5 6</b>
10. I meet my own personal expectations for expertise in caring for my child.	<b>1 2 3 4 5 6</b>
11. If anyone can find the answer to what is troubling my child, I am the one.	<b>1 2 3 4 5 6</b>
12. My talents and interests are in other areas, not in being a parent.	<b>1 2 3 4 5 6</b>
13. Considering how long I've been a mother/father, I feel thoroughly familiar with this role.	<b>1 2 3 4 5 6</b>
14. If being a mother/father of a child were only <u>more</u> interesting, I would be motivated to do a better job as a parent.	<b>1 2 3 4 5 6</b>
15. I honestly believe I have all the skills necessary to be a good mother/father to my child.	<b>1 2 3 4 5 6</b>
16. Being a parent makes me tense and anxious.	<b>1 2 3 4 5 6</b>
17. Being a good mother/father is a reward in itself.	<b>1 2 3 4 5 6</b>

## Appendix 9: The Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True (A), Somewhat True (B), or Certainly True (C). It would help us if you answered all the items as best you can even if you are not absolutely certain or the items seem daft! Please give your answers on the basis of the child's behaviour over the last six months.

A	B	C
Not True	Somewhat True	Certainly True
		Considerate of other people's feelings.
		Restless, overactive, cannot stay still for long.
		Often complains of headaches, stomach-aches, or sickness.
		Shares readily with other children (treats, toys, pencils etc.)
		Often has temper tantrums or hot tempers
		Rather solitary, tends to play alone.
		Generally obedient, usually does what adults request
		Many worries, often seems worried
		Helpful if someone is hurt, upset, or feeling ill.
		Constantly fidgeting or squirming.
		Has at least one good friend.
		Often fights with other children or bullies them.
		Often unhappy, down-hearted, or tearful.
		Generally liked by other children.
		Easily distracted, concentration wanders.
		Nervous or clingy in new situations, easily loses confidence.
		Kind to younger children.
		Often lies or cheats.
		Picked on or bullied by other children.
		Often volunteers to help others (parents, teachers, other children).
		Thinks things out before acting.
		Steals from home, school or elsewhere.
		Gets on better with adults than other children.
		Many fears, easily scared.
		Sees tasks through to the end, good attention span.

**Do you have any other comments or concerns?**



## **Appendix 10: The Social Communication Questionnaire**

**Not included due to copyright restrictions**

## Appendix 11: Demographics Questionnaire

While it is helpful if you answer all questions, if there are any questions you would prefer not to answer please leave these questions blank.

Age \_\_\_\_\_ Gender \_\_\_\_\_

Ethnicity \_\_\_\_\_

Employment Status \_\_\_\_\_

Hours worked during the week \_\_\_\_\_

Highest Education *(please circle your answer)*

Age 16 or below      A-levels      Degree/diploma      Post-graduate

Marital Status

Married Single      Divorced      Co-Habiting      Separated

Number of children \_\_\_\_\_

Relationship to child

Biological parent    Step-parent    Adopted parent    Biological parent's partner    Other

Do you live with the child?      Yes      No

Gender of Child      Male      Female

**Birth Order of Child**

Oldest

Youngest

Middle

Only child

**How long is it since your child received a neurodevelopmental diagnosis?**

*(A neurodevelopmental disorder is one in which there is an impairment of the growth and development of the [brain](#) or [central nervous system](#).) (Please circle.)*

Less than 1 month    1-3 months    4-6 months    6-12 months    1-2 years    +2  
years

My child has never received a neurodevelopmental diagnosis

**How long is it since your child received a paediatric (medical) diagnosis?**

*(A paediatric/medical diagnosis of any physical disease, disorder or impairment not classed as a neurodevelopmental disorder.) (Please circle.)*

Less than 1 month    1-3 months    4-6 months    6-12 months    1-2 years    +2  
years

My child has never received a medical diagnosis

## Appendix 12: PUN-Q Inter-Item Correlation Matrices

**Appendix 12a:** Correlation matrix of Time 1 PUN-Q-8

	Item 1	Item 2	Item 3	Item 4	Item 5	Item 6	Item 7	Item 8
<b>Item 1</b>	1.00							
<b>Item 2</b>	.57	1.00						
<b>Item 3</b>	.51	.46	1.00					
<b>Item 4</b>	.19	.39	.32	1.00				
<b>Item 5</b>	.13	.36	.11	.26	1.00			
<b>Item 6</b>	.33	.22	.37	.20	.23	1.00		
<b>Item 7</b>	-.03	.26	-.10	.22	.12	-.09	1.00	
<b>Item 8</b>	-.15	.21	.14	.51	.11	.31	.20	1.00
<b>N correlations&gt;.90</b>	0	0	0	0	0	0	0	0

**Appendix 12b:** Correlation matrix of Time 2 PUN-Q-8

	Item 1	Item 2	Item 3	Item 4	Item 5	Item 6	Item 7	Item 8
<b>Item 1</b>	1.00							
<b>Item 2</b>	.45	1.00						
<b>Item 3</b>	.48	.39	1.00					
<b>Item 4</b>	.39	.32	.38	1.00				
<b>Item 5</b>	.21	.44	.24	.17	1.00			
<b>Item 6</b>	.39	.08	.32	.37	-.04	1.00		
<b>Item 7</b>	.08	.24	.37	.25	.23	.07	1.00	
<b>Item 8</b>	.07	.23	.45	.52	.30	.24	.55	1.00
<b>N correlations&gt;.90</b>	0	0	0	0	0	0	0	0

**Appendix 12c:** Correlation matrix of Time 3 PUN-Q-13

	Item 1	Item 2	Item 3	Item 4	Item 5	Item 6	Item 7	Item 8	Item 9	Item 10	Item 11	Item 12	Item 13
<b>Item 1</b>	1.00												
<b>Item 2</b>	.67	1.00											
<b>Item 3</b>	.61	.47	1.00										
<b>Item 4</b>	.38	.42	-.12	1.00									
<b>Item 5</b>	.74	.45	.43	.66	1.00								
<b>Item 6</b>	.65	.71	.47	.27	.37	1.00							
<b>Item 7</b>	.79	.70	.44	.56	.59	.62	1.00						
<b>Item 8</b>	.29	.13	-.32	.47	.24	.21	.10	1.00					
<b>Item 9</b>	.31	.08	.45	.22	.16	.25	.27	-.06	1.00				
<b>Item 10</b>	.55	.46	.18	.83	.91	.25	.58	.33	.02	1.00			
<b>Item 11</b>	.44	.42	.15	.54	.61	.19	.50	.26	-.12	.80	1.00		
<b>Item 12</b>	.60	.43	.01	.93	.78	.41	.69	.55	.24	.86	.64	1.00	
<b>Item 13</b>	.65	.67	.08	.79	.60	.52	.68	.44	.34	.67	.63	.84	1.00
<b>N</b>	0	0	0	1	1	0	0	0	0	0	0	0	0
<b>correlations&gt;.90</b>													